ANNALS OF SURGERY

VOL. 127 MARCH, 1948 No. 3

COMPLETE TRANSPOSITION OF THE AORTA AND THE PULMONARY ARTERY*

Experimental Observations on Venous Shunts as Corrective Procedures.

C. ROLLINS HANLON, M.D., AND ALFRED BLALOCK, M.D.

BALTIMORE, MD.

FROM THE DEPARTMENT OF SURGERY OF THE JOHNS HOPKINS UNIVERSITY AND THE JOHNS HOPKINS HOSPITAL

COMPLETE TRANSPOSITION of the aorta and the pulmonary artery is an infrequently encountered condition[1, 13, 15, 16] which may be diagnosed with fair accuracy during life.[17] Recent progress in the surgical therapy of other types of congenital heart disease has prompted us to approach the problem of transposition in a similar fashion. We shall discuss some possible modes of surgical attack on transposition of the great vessels after a brief review of its pathologic anatomy and physiology.

The pathogenesis of transposition is still not wholly clear despite intensive study of the problem for more than a century. A number of ingenious and conflicting theories have been adduced, none of which is completely satisfactory, and the accompanying differences in terminology have further confused the issue. For a critical review of the subject one may consult the recent papers of Harris and Farber[6] and Lev and Saphir.[10]

In complete ("uncorrected") transposition of the great vessels the aorta arises from the ventricle receiving systemic venous blood and the pulmonary artery arises from the ventricle receiving oxygenated blood. Blood pumped out by the left ventricle through the pulmonary artery to the lungs returns through the pulmonary veins and left auricle to its point of origin; the right ventricular blood emerges through the aorta and proceeds in a similarly closed circuit through the systemic circulation. The greater and lesser circulations are thus basically separate, a condition obviously incompatible with continued existence. In most instances, however, some degree of communication exists between the two circulations by way of septal defects or other abnormalities. These compensating abnormalities send some oxygenated blood through the systemic circulation, thus permitting the patients to survive for variable periods.

"Transposition" in pure form consists merely of a posterior origin of the pulmonary artery that goes directly back to the lungs, whereas the aorta arises anteriorly and escapes its usual encirclement by the pulmonary artery. If the ventricles are similarly reversed in position no physiologic abnormality

* Aided by a grant from The Carolyn Rose Strauss Foundation, Monroe, Louisiana.

is evident and the transposition is "corrected." But if the pulmonary artery arises from the ventricle receiving oxygenated pulmonary blood and the aorta from the ventricle receiving systemic venous blood, the transposition is termed "complete," "uncorrected," or "transpositio vera." It is this latter condition which we shall discuss.

The term "uncorrected" was adopted presumably to indicate that the abnormal position of the great vessels was not "corrected" by a similar change in the position of the ventricles. It is a poor term, however, since it suggests that no other compensatory abnormality is present, such as interatrial or interventricular septal defects. One must realize, however, that every patient with "uncorrected" transposition who survives beyond fetal life has additional abnormalities tending to correct the lack of mixing between the two circulations. Because of this confusion we favor the term "complete transposition" ("transpositio vera").

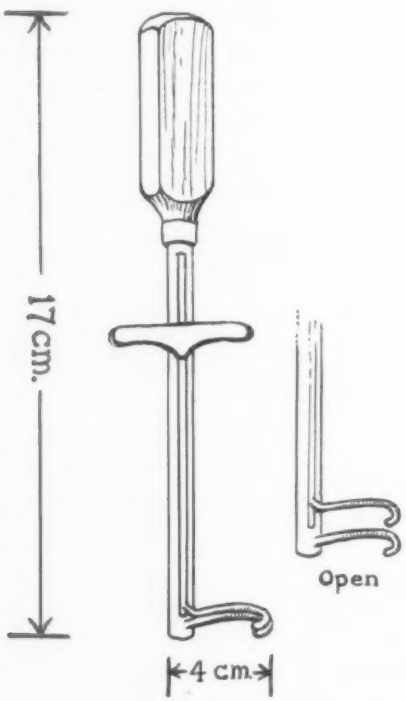


FIG. 1.—Special clamp for end-to-side anastomosis.

For a patient with complete transposition to survive after birth there must be a means by which oxygenated blood reaches the aorta. This may be effected by way of an interatrial or interventricular septal defect, by a patent ductus arteriosus, or by passage of pulmonary venous blood into either vena cava. Such abnormalities in various combinations provide a degree of compensation which in rare instances has resulted in prolonged and fairly normal existence.

The literature on this point has been surveyed by Kato,[8] who collected 92 cases of transpositio vera and added five of his own. In 85 of these 97 cases the data are sufficient for analysis. At least 15 cases have since been reported by others,[5, 6, 10, 11, 12, 14, 16, 18] and we include an additional 23 studied at autopsy in The Johns Hopkins Hospital to make a total of 123 complete transpositions.

Of Kato's 85 cases, 70 died in less than a year. In those with complete transposition and an atrial septal defect the survival time averaged over two years, while in those with transposition and a ventricular septal defect the average age at death was well over four years. The combination of interatrial and interventricular septal defects gave an average survival time of five and one-half years in 16 cases. It should be noted that the inclusion of two individuals aged 19 and 56 years lends an unduly favorable character to the

average survival time for the group with combined defects. Without this long-surviving pair the average life span of the group would be less than a year. Long survivals are of course quite likely to be reported individually in the literature, while stillbirths or neonatal mortalities are included only in large series or not reported at all.

In the 123 collected cases the average duration of life was nineteen months. Six patients lived ten years or longer; if these are excluded, the average duration of life for 117 patients is only five and one-half months. The survival time of patients with complete transposition and various associated abnormalities is seen in the following table:

TABLE I.—*Complete Transposition: Duration of Life with Various Associated Abnormalities*

Associated Abnormality	No. of Cases	Duration of Life
Patent foramen ovale*	13	1 yr. 11 mo.
Patent ductus arteriosus	12	64 days
Patent foramen ovale plus ductus arteriosus	44	40 days
Interventricular septal defect	12	4 yrs. 1 mo.
Patent foramen ovale plus interventricular septal defect	19	4 yrs. 9 mo.
Patent ductus arteriosus plus interventricular septal defect	7	2 yrs. 3 mo.
Patent foramen ovale and ductus arteriosus plus interventricular septal defect	16	5 mo.
Total	123	Average duration—19 mo.

* Indicates any interatrial septal defect.

It is apparent that in complete transposition an interventricular septal defect is the single compensating abnormality associated with the largest life expectancy. A patent interatrial septum is the next most favorable isolated defect, and the combination of these two gives the best prognosis of all.

As a result of these defects some oxygenated blood passes into the aorta. A similar result may occur when pulmonary veins empty into the systemic venous auricle or into either vena cava.

Surgical therapy for transposition of the great vessels might reproduce any of the defects mentioned above. Interventricular defects have been produced experimentally for many years, but it is difficult to control the size of the defect, and a blind approach through the ventricular wall makes the procedure both uncertain and hazardous. Further developments in the technic of intracardiac manipulation under cardioscopic control may alter this aspect of the problem, but our current discussion is limited to experimental procedures carried out under direct vision.

Having excluded interventricular septal defects and leaving the technic of producing interatrial defects and other procedures to subsequent communications, we may consider the artificial production of pulmonary venous return to the right side of the heart. This condition is found occasionally at autopsy in patients who have shown no symptoms of cardiovascular abnormality. The number of pulmonary veins entering the right side varies from one vein up to

all the veins, a condition which is found in about a third of these individuals. The subject has been well reviewed by Brody,[3] and the surgical significance of anomalous pulmonary veins has recently been stressed by Brantigan.[2] This author mentions the possibility of transferring aberrant pulmonary veins from the right atrium or its tributaries into the left atrium. In the present studies we are concerned with a directly opposite procedure, namely, the anastomosis of pulmonary veins to the right atrium or its tributaries.

Such a venous shunt would bring oxygenated blood to the right side of the heart. To test the effect of this on an experimental transposition, one must first produce the corrective shunt and subsequently produce the transposition; if the sequence of the procedures were reversed the animal could not possibly survive. In a number of attempts, which are being continued, we have not yet succeeded in producing a complete transposition of the aorta and the pulmonary artery in the dog. Therefore our observations are confined to: (1) anastomoses of the pulmonary veins to the right auricle, and (2) anastomoses of the pulmonary veins to the superior vena cava.

METHODS AND PROCEDURES

Healthy adult mongrel dogs were used, varying in weight from 6.0 to 20.0 kg. The average weight was 10.6 kg. After premedication with morphine sulfate (¼ to ½ gr.) and $\frac{1}{150}$ gr. of atropine sulfate they were anesthetized by drop ether followed by introduction of an endotracheal catheter with inflatable cuff. A mechanical device provided regular periodic inflation of the lungs with ether vapor while the chest was open. An intercostal approach through the right fourth interspace afforded adequate exposure in all cases. At the conclusion of the procedure the ribs were approximated by absorbable pericostal sutures and air was evacuated from the pleural cavity before closure of the skin by means of an intercostal catheter. All wounds were closed in layers with fine silk.

In both groups of animals we used a special occlusive clamp which allowed bloodless access to one portion of the heart and veins while permitting a relatively free flow of blood in their remaining portions. This clamp (Fig. 1) consists essentially of two semicircular jaws of rounded wire, the upper one sliding in a grooved handle and capable of exerting pressure against the lower, fixed jaw by means of a spring.

I. Anastomosis of Pulmonary Veins to the Right Auricle

The veins from the apical and cardiac lobes of the right lung are isolated and divided after ligation at their point of entry into the left auricle. This involves sharp dissection in the plane between the ventral wall of the pulmonary veins and the adherent dorsal wall of the right auricle. It is usually necessary to free the veins for a full centimeter medial to their point of attachment to the right auricle. This allows one to work with a good length of common pulmonary vein. Backflow is prevented by bulldog clamps on the tributary veins from the two lobes.

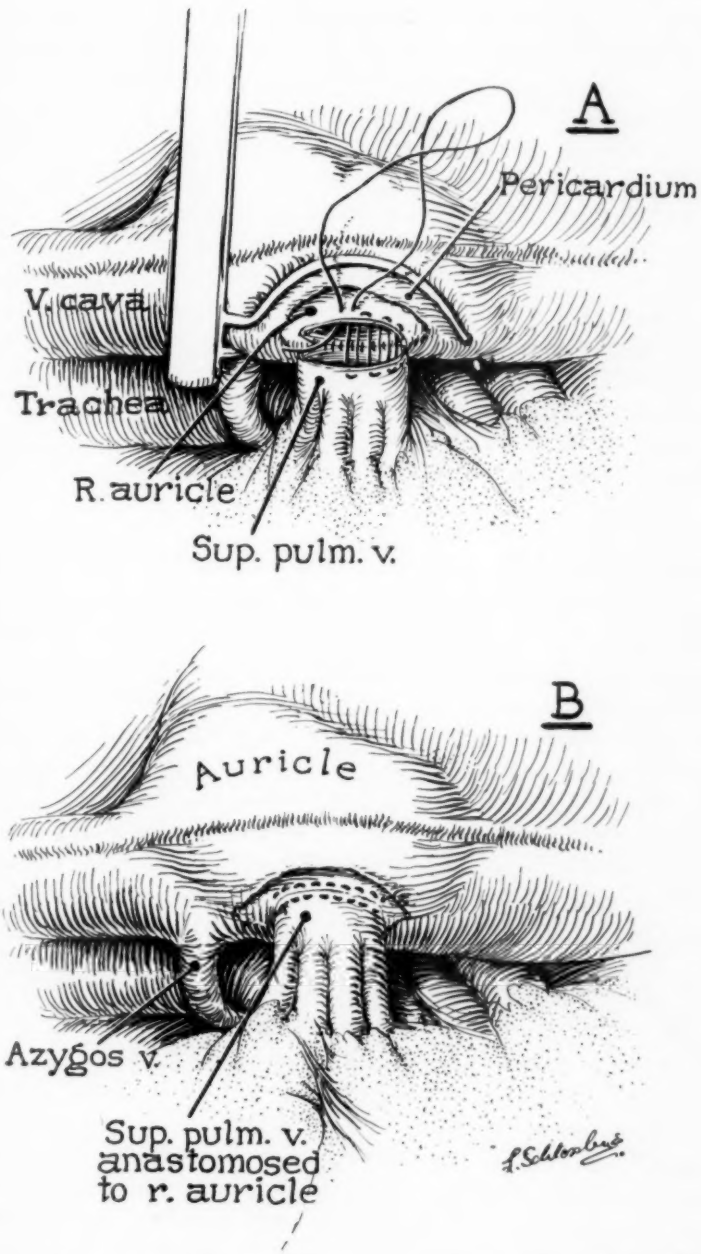


FIG. 2.—(A) With occlusive clamp preventing bleeding from the right auricle, a circular anastomosis is being made between the superior pulmonary veins and the right auricle. Bulldog clamps preventing backflow from the lung are omitted from the drawings for the sake of clarity.
(B) Completed anastomosis.

The pericardium is reflected from the right auricle and the spring clamp is applied to the ventral and lateral aspect of the auricle, thus isolating a crescentic segment. This portion of the auricle is seen externally as the point of junction between the two cavae, from which it differs little in gross appearance (Fig. 2A). Histologically, however, it is seen to resemble auricular musculature, without the characteristic trabeculations. An elliptical portion of the auricle is excised with scissors to give an opening comparable in size to the lumen of the superior pulmonary veins, usually 10 to 15 mm. in diameter. Into this auricular opening the superior pulmonary veins are implanted by a running mattress suture which everts the intima and is interrupted at several points by everting mattress stay sutures (Fig. 2B). On removal of the clamp there may be a temporary oozing of blood along the suture line, readily controlled by a short period of light pressure. Additional sutures are seldom required. A few fine sutures close the pericardium over the anastomosis. The branches of the pulmonary artery to the upper lobes are occluded during the performance of this anastomosis as well as in the procedure described below.

II. Anastomosis of Pulmonary Veins to the Superior Vena Cava

This procedure closely resembles the one just described, except that the veins are implanted into a defect formed by excision of the azygos vein at its point of entry into the superior vena cava. It is usually necessary to free the pericardium from the vena cava and to expose the vessel thoroughly before application of the spring clamp. With the clamp in place there need be no impairment of venous return to the right auricle by way of the superior vena cava (Fig. 3A). The azygos vein and a circular segment of the wall of the vena cava are excised with scissors, giving an opening comparable in size to that of the pulmonary veins. A circular anastomosis is carried out as previously described, and the clamp is removed, allowing entry of oxygenated blood to the right auricle by way of the superior vena cava (Fig. 3B).

RESULTS

All of the operative procedures were well tolerated, except for one death due to anesthesia. At varying periods after operation the patency of the anastomosis was investigated by angiocardiography or by direct catheterization of the right superior pulmonary veins through the jugular vein.

The animals were killed under anesthesia at intervals up to one hundred and one days. In some instances the condition of the anastomosis was investigated roentgenologically after injection of Hill's mass, and in nearly all cases the autopsy included microscopic examination as well as gross study.

Pulmonary Veins Into Auricle. In ten instances the pulmonary veins from the upper lobes of the right lung were anastomosed into the right auricle by the technic described. There were no operative deaths. Physiologic studies demonstrated the patency of the anastomosis in some cases; when patency could not be shown, autopsy usually confirmed the obliteration

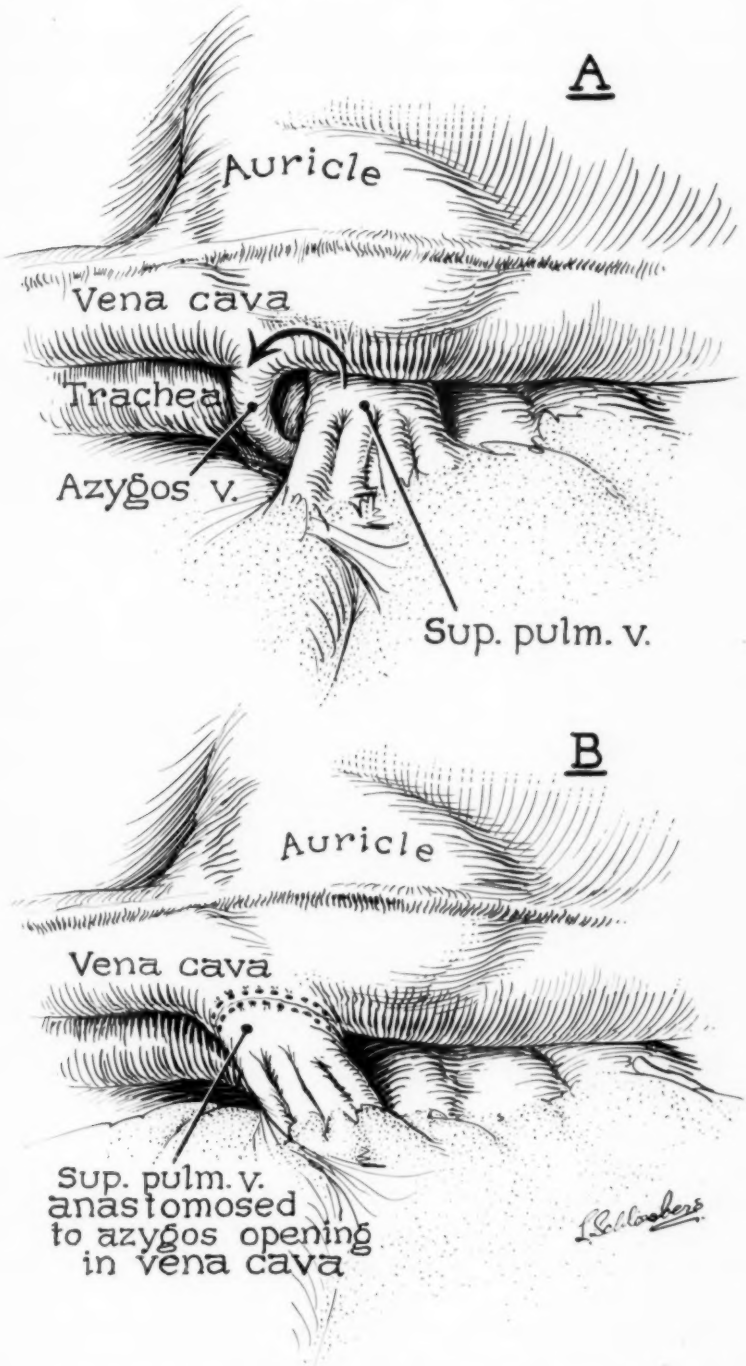


FIG. 3.—(A) Arrow indicates proposed anastomosis of the superior pulmonary veins to the defect left by excision of a segment of superior vena cava at the azygos opening.

(B) Completed anastomosis.

of the stoma. In some instances marked constriction of the stoma rendered catheterization impossible, even though an opening several millimeters in diameter remained.

When difficulty had been encountered in catheterization of the pulmonary vein one commonly saw subendocardial hemorrhages in the auricle at autopsy. This observation suggested that vigorous attempts at catheterization might

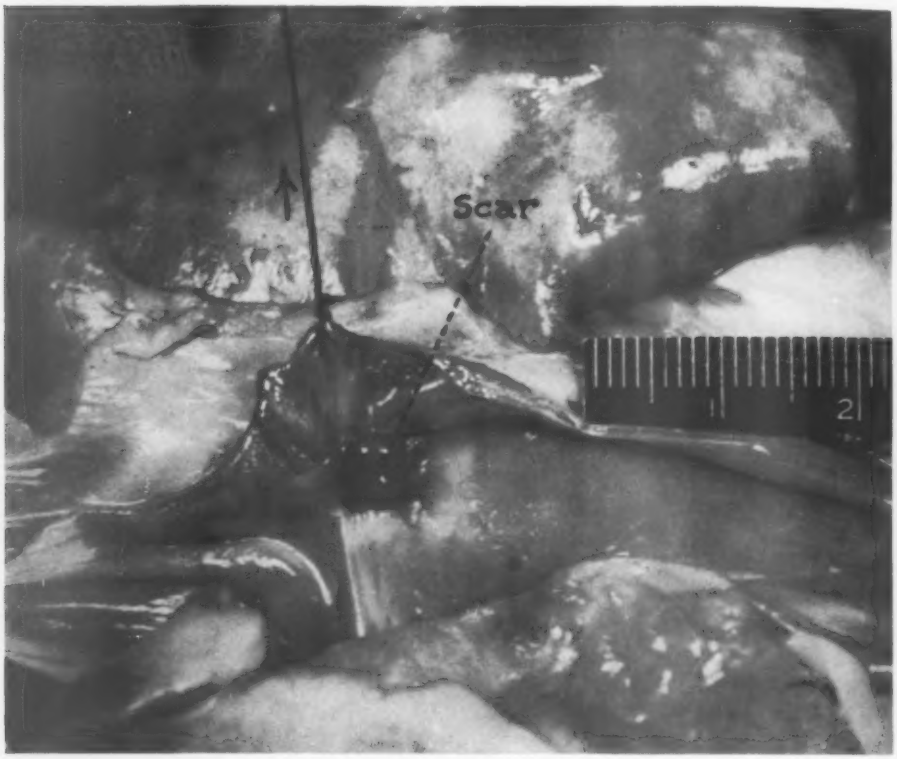


FIG. 4.—Right auricle opened to show hyperplastic scar at the site of an unsuccessful anastomosis.

contribute to closure of the stoma by scarring. The site of an obliterated anastomosis was generally represented by a hyperplastic scar, quite readily detected during life by palpation with the rigid cannula passed through the jugular vein. The appearance of such a scar may be seen in the accompanying photograph (Fig. 4).

In contrast to the appearance in Figure 4 is the appearance of a successful result shown in Figure 5. One may see the smoothly healed, oval anastomosis as well as the openings of the pulmonary veins. The microscopic appearance of the junction of the thin-walled veins with the relatively thick auricular muscle is seen in Figure 6.

Results were evaluated by gross and microscopic examination of the operative site from 26 to 101 days after operation. Four of the results were satisfactory, six were unsatisfactory. In three instances the anastomosis was widely patent, in one there was some constriction of the opening, in three the constriction was quite pronounced, and three were totally occluded.

Technical difficulties were encountered while the procedure was being developed, and it was in these early experiments that the unsatisfactory results were largely obtained. It must be noted, however, that even in the final operation in this series some constriction of the anastomosis was found

FIG. 5.—Excellent result 26 days after anastomosis of superior pulmonary veins to the right auricle. The openings of the tributary veins are well demonstrated.

at autopsy. This finding suggests that technical difficulties are inherent in the operation, and it seems likely that the thick auricular wall, especially in its trabeculated area, is not well suited to vascular anastomosis.

Pulmonary Veins to Superior Vena Cava. The pulmonary veins were anastomosed to the superior vena cava at the azygos opening in 15 animals. There were no deaths during the operative procedure. Two animals died of infection, four and eight days after operation. These two anastomoses were widely patent at the time of death, but of course insufficient time had elapsed to evaluate the result.

In the remaining 13 dogs there were 10 excellent results, two with a patent but constricted anastomosis, and one in which the anastomosis was totally occluded.

In all three preparations showing poor results some technical mishap occurred during the performance of the anastomosis. In one instance a suture placed to control a tear in the pulmonary vein had completely occluded the anastomosis.

The gross and microscopic picture in a case that healed well may be seen in the accompanying illustrations. The site of anastomosis is viewed from the interior of the superior vena cava in a dog killed after 83 days (Fig. 7).

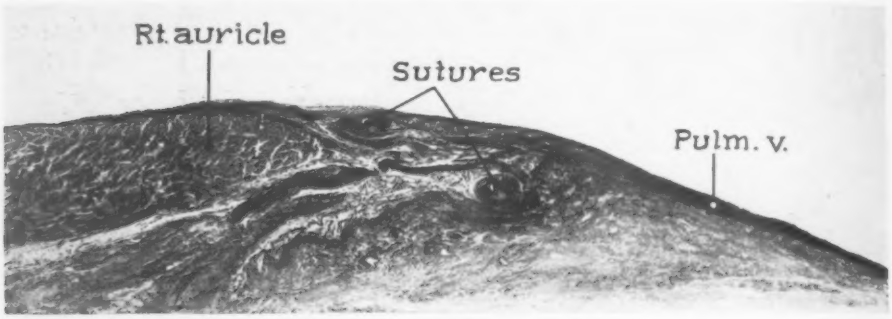


FIG. 6.—Junction of pulmonary veins and right auricle showing disparity in thickness of walls. (30x) (Illustration retouched)

Healing is smooth with negligible scarring. The photomicrograph shows the junction of the pulmonary vein and the superior vena cava and illustrates well the similarity in thickness of the vessel walls (Fig. 8). Such similarity is in striking contrast to the disparity in thickness of the wall of the pulmonary vein and the right auricle (Fig. 6). As previously noted, suture of this thick auricular muscle is frequently accompanied by scarring and obliteration of the anastomosis.

DISCUSSION

The basic difficulty in complete transposition of the aorta and the pulmonary artery is the failure of oxygenated blood to reach the ventricle which empties into the aorta. Survival of such patients is directly related to the degree of interchange between the two sides of the heart by way of persistent fetal passages. Patients with a complete transposition and a patent foramen ovale have lived as long as ten years, and those with a large interventricular defect as a compensatory shunt have lived up to 21 years. A combination of these two compensatory abnormalities allowed one patient

with a complete transposition to live an essentially normal life for 56 years. However, the average duration of life in a total of 123 reported cases of complete transposition is only 19 months.

The return of pulmonary vein blood into the vena cava or the right auricle provides another possible means of compensation in complete transposition. This condition is occasionally detected as an isolated abnormality at operation or autopsy in patients who have shown no clinical evidence of cardiovascular difficulty. It has also been described as a beneficial adjustment in transposi-

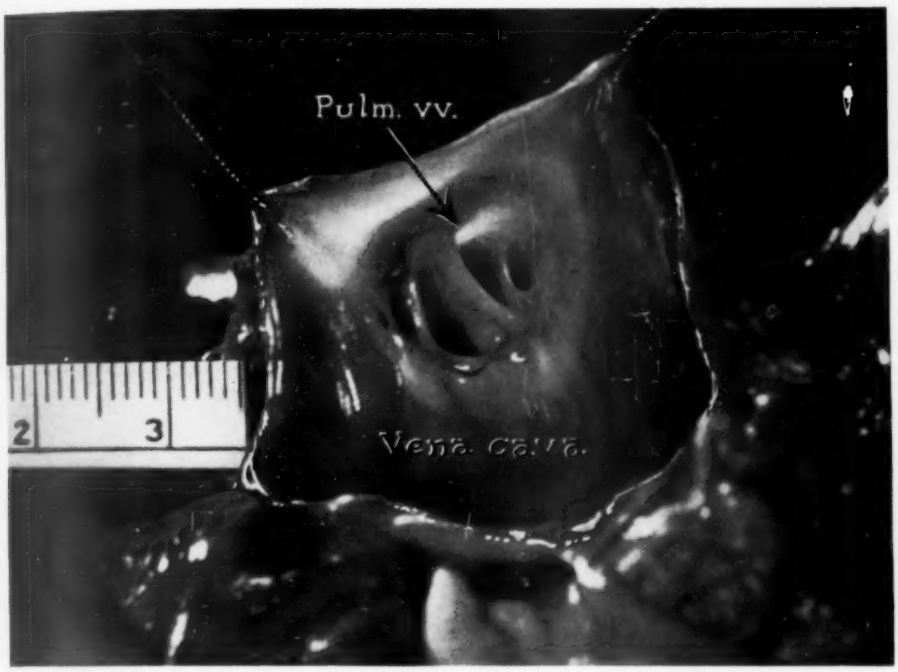


FIG. 7.—Vena cava opened to show smooth healing 83 days after anastomosis of superior pulmonary veins to vena cava.

tion. The possibility of its use in the surgical treatment of transposition prompted the studies detailed here.

These studies consisted of anastomoses in dogs of the veins from the two upper lobes of the right lung to the superior vena cava or the right auricle. In general, the results of anastomosis to the superior vena cava were excellent whereas anastomoses to the auricle tended to become occluded. It seems likely that the thick auricular wall is poorly adapted to vascular anastomosis.

The ability of such anastomoses to return blood to the right side of the heart has been established by catheterization of the superior vena cava and the cardiac chambers. In order to test the therapeutic effectiveness of these anastomoses, however, one must observe their effect on an experimental

animal with complete transposition of the arterial trunks. Since a complete transposition as such would be promptly fatal, it is necessary to establish the corrective pulmonary venous shunt before producing the transposition. Despite a number of efforts which are being continued, we have not yet succeeded in preparing an animal in which a complete transposition of the arterial trunks is corrected sufficiently by artificial shunts to permit life for more than a few minutes.

Despite the failure to reproduce complete transposition in the experimental animal and the consequent inability to test the therapeutic effect of the different shunting procedures, these experiments demonstrate that oxygenated blood may be transmitted to the right side of the heart by way of the transplanted pulmonary veins. The anastomoses to the superior vena cava remained patent up to three and a half months in more than three-fourths of the animals subjected to operation; technical difficulties at operation accounted for partial or complete closure in the remaining animals.

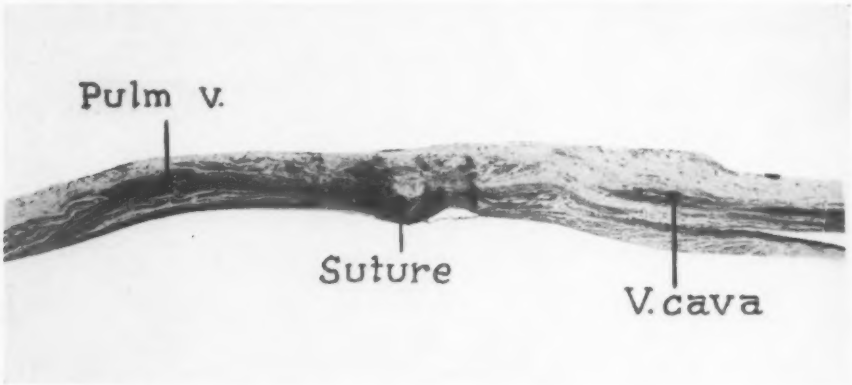


FIG. 8.—Junction of pulmonary veins and vena cava 30 days after operation. (10x)

SUMMARY

In complete transposition of the aorta and the pulmonary artery in man, the time of survival is dependent on the degree of mixing between the greater and the lesser circulations.

Such mixing may be effected by interatrial or interventricular septal defects, by a patent ductus arteriosus, or by entry of pulmonary veins into the transposed right side of the heart. The literature on this point has been briefly reviewed.

The technical feasibility of anastomosing the pulmonary veins to the right auricle or to the superior vena cava has been investigated in the dog.

Anastomosis of the pulmonary veins to the right auricle was usually unsatisfactory whereas an anastomosis of the pulmonary veins to the superior vena cava remained patent in over three-fourths of the experiments.

Anastomosis of the pulmonary veins to the superior vena cava appears

feasible in man and offers one possible approach to the surgical treatment of complete transposition of the great cardiac arteries.

BIBLIOGRAPHY

1 Abbott, M. E.: Atlas of Congenital Heart Disease. New York, American Heart Association, 1936.

2 Brantigan, O. C.: Anomalies of the Pulmonary Veins—Their Surgical Significance. Surg. Gynec. and Obstet., **84**: 653, 1947.

3 Brody, H.: Drainage of the Pulmonary Veins into the Right Side of the Heart. Arch. path., **33**: 221, 1942.

4 Doerr, W.: Zur Transposition der Herzschlagadern. Virchows Arch. f. path. anat., **303**: 168, 1938–1939.

5 Feldman, W., and A. Chalmers: A Case of Complete Transposition of the Great Vessels of the Heart with a Patent Foramen Ovale. Brit. J. Child. Dis., **30**: 27, 1933.

6 Harris, J. S., and S. Farber: Transposition of the Great Cardiac Vessels with Special Reference to the Phylogenetic Theory of Spitzer. Arch. path., **28**: 427, 1939.

7 Ingham, D. W., and F. A. Willius: Congenital Transposition of the Great Arterial Trunks. Am. Heart J., **15**: 482, 1938.

8 Kato, Katsuji: Congenital Transposition of Cardiac Vessels. A Clinical and Pathologic Study. Am. J. Dis. Child., **39**: 363, 1930.

9 Lev, Maurice, and Otto Saphir: Transposition of Large Vessels. J. Tech. Methods, **17**: 126, 1937.

10 *Idem:* A Theory of Transposition of the Arterial Trunks Based on the Phylogenetic and Ontogenetic Development of the Heart. Arch. path., **39**: 172, 1945.

11 Lia, Bianca R., and Olga C. Leary: Complete Transposition of the Aorta and Pulmonary Artery. Am. Heart J., **18**: 108, 1939.

12 Moore, Robert A.: Transposition of the Great Arterial Trunks (Report of a Case). J. Tech. Methods, **12**: 184, 1929.

13 Rannels, Herman W., and James H. Propst: Incidence of Congenital Cardiac Anomalies found at Autopsies Performed in the Hospital of the University of Pennsylvania. J. Tech. Methods, **17**: 113, 1937.

14 Read, Wm. T., and E. B. Krumbhaar: Eight Cases of Congenital Heart Disease. Med. Clin. N. Amer., **16**: 229, 1932.

15 Roberts, Joseph T.: Incidence of Congenital Heart Disease in Charity Hospital of New Orleans. J. Tech. Methods, **17**: 108, 1937.

16 Szypulski, John T.: Study of Congenital Heart Disease at Philadelphia General Hospital. J. Tech. Methods, **17**: 119, 1937.

17 Taussig, H. B.: Complete Transposition of the Great Vessels. Clinical and Pathologic Features. Am. Heart J., **16**: 728, 1938.

18 Walker, J. W., and V. J. Dardinski: Complete Uncorrected Transposition of the Vessels. Report of a Case. Arch. Pediat., **62**: 209, 1945.

THE SIGMOID AS A SOURCE OF RIGHT-SIDED SYMPTOMS*

ALBERT S. LYONS, M.D., F.A.C.S.
NEW YORK, N. Y.

* FROM THE SURGICAL SERVICES OF THE MOUNT SINAI HOSPITAL, NEW YORK, N. Y.

MANY SURGEONS have had the experience of operating upon a patient with symptoms resembling acute appendicitis, only to find a sigmoidal lesion responsible for the clinical signs. Such experience, however, has been reported only as an incidental finding and the frequency of occurrence of left-sided lesions with right-sided symptoms has not generally been recognized. The principal pathological conditions of the colon responsible for the confusion of signs are diverticulitis and carcinoma, especially with perforation.

That the right-sided syndrome is not infrequent is seen from an analysis of the many individual reports of cases and of the summaries dealing with diverticulitis and carcinoma. For example, Allen[1] reported seven cases of carcinoma of the colon to demonstrate the high mortality following perforation. However, his descriptions indicate that of the five sigmoidal cases, three had the chief symptoms and signs on the right side of the abdomen, and in the fourth case the symptoms began on the right, though they later shifted to the left. Examination of a review of diverticulitis by Arnheim[2] discloses that of the 13 cases with peritonitis, three were operated upon with the diagnosis of acute appendicitis and a fourth case, though known to have diverticulitis, had right-sided symptoms and signs. Young and Young[3] summarized statistically 84 cases of diverticulitis of the colon. Their report contains three chronic cases and nine acute cases with symptoms or signs on the right. Some of the patients of Cave and Alsop[4] who had perforations of inflamed diverticula of the sigmoid were operated upon with a diagnosis of acute appendicitis, and a right McBurney incision was used in some instances. In the series reported by Morton,[5] pain was present in the right lower quadrant in five instances and tenderness was found occasionally on the right in cases with perforation. Rankin and Grimes[6] have stated that it is common for acute perforative diverticulitis to present atypical symptoms of acute appendicitis and that the surgeon will often use a right McBurney or right rectus incision. Jones[7] described a particularly striking case with acute symptoms resembling appendicitis and a palpable mass on the right side of the abdomen which was drained as an appendiceal abscess but was later found to have been the result of a perforated sigmoidal diverticulum. Yet other monographs on diverticulitis,[8, 9, 10, 11] and on surgery of the colon,[12, 13] as well as the standard textbooks[14, 15, 16] and numerous papers[17, 18, 19, 20, 21] on diverticulitis and carcinoma of the colon, fail to mention the relative frequency of this crossed symptomatology. Even Rankin, Bargen, and Buie,[22] while stating that they have observed four cases of diverticulitis in which pain was referred to the right side, further observe that "this is extremely unusual."

The reasons for the referral of pain to the opposite side of the abdomen become clear when one realizes that the sigmoid and rectosigmoid are not always left sided in anatomical position. Roentgenograms of the colon frequently show a rectosigmoid and lower sigmoid to the right of the midline. Lynch[23] suggested that mobility of the lower sigmoid could account for right-sided symptoms resembling appendicitis in cases with diverticulitis. Similarly, Graham[24] stated, "Because of the mobility of the sigmoid, it may be in either right or left iliac fossa." Morton[5] recently mentioned the fact that at operation the sigmoid may be found lying toward the right. Laufman[25] and Jones[7] noted that the sigmoid is very frequently in the midline or on the right side. Moreover, extensive cadaver dissections by Ssoson-Jaroschewitsch[26] showed that approximately 11 per cent of sigmoids have a course which is chiefly on the right side of the abdomen.

An additional factor, either in association with other causes or acting independently, may be distention of the cecum. Either organic stenosis or colonic spasm alone can produce intestinal obstruction. Colp[27] reviewed all the previous reports and described five additional instances of colonic obstruction produced by spasm. Moreover, in the presence of diverticulitis and sometimes of carcinoma, spasm of the sigmoid without actual obstruction is frequently so marked that the back pressure created in the colon may be focussed chiefly in the cecum. Kantor[28] called attention in this country to the occurrence of ulcerations in the cecum due to marked colonic distention, and he further indicated[29] that pain in the cecal region could be produced by lesions causing spasm in the distal part of the colon. Although the symptomatology of the cases herein presented was not produced exclusively by cecal distention, this mechanism may have been a contributing factor in some instances.

In the present study 18 cases are reported with lesions in the sigmoid and rectosigmoid which caused right-sided symptoms and signs. In 14 the reference of pain and tenderness to the right was marked enough to prompt operative intervention through incisions placed on the right side of the abdomen.

These cases are used to illustrate the probable causes of the right-sided symptoms: 1. Sigmoid lying over on the right of the midline; 2. Perforation of the right wall of the rectosigmoid with spillage of fluid toward the right; 3. Perisigmoidal abscess extending to the right; 4. Adherence of right-sided structures to the sigmoidal lesion; 5. Situs inversus.

SIGMOID LYING ON THE RIGHT

Case 1: (Admission No. 480580) J. F., a 58-year-old man, was admitted to The Mount Sinai Hospital on October 18, 1941, with a history of lower abdominal pain for one day, shifting shortly after onset to the right lower quadrant.

On physical examination, temperature was 101. There was tenderness in both lower quadrants of the abdomen, most marked on the right, with rebound tenderness on the right.

The preoperative diagnosis was acute appendicitis.

Operation was performed on October 18, 1941, through a right McBurney incision. A loop of redundant sigmoid lay on the right side of the abdomen. The wall was markedly reddened and swollen over an area about one-half inch in diameter at the attachment of

the mesentery, which was also edematous. Flakes of fibrin covered this localized, reddened area. A small amount of free, cloudy, peritoneal fluid was present. The appendix was normal. The sigmoid area was drained.

The final diagnosis was acute diverticulitis.

Case 2: (Admission No. 526864) M. K., a 52-year-old man, was admitted to The Mount Sinai Hospital on November 1, 1944. In the past he had had intermittent attacks of right lower quadrant pain. Two days before admission he had severe lower abdominal pain, associated with nausea and fever.

Physical examination showed a temperature of 100. There was marked tenderness and spasm in the right lower quadrant, especially over McBurney's point.

The preoperative diagnosis was acute appendicitis.

Operation on November 1, 1944 was performed through a right McBurney incision. The appendix, which pointed medially, seemed mildly inflamed by contiguity with a markedly inflamed sigmoid which lay near the appendix. The appendix was removed. Pathological report showed it to be normal.

The final diagnosis was acute diverticulitis.

Case 3: (Admission No. 446923) G. H., a 56-year-old man, had chronic constipation for many years. Six days before admission the constipation became more marked. Six hours before admission he had severe lower abdominal pain, most marked on the right. He was admitted to The Mount Sinai Hospital on October 7, 1939.

On physical examination, the temperature was 99.6. There was diffuse abdominal tenderness, most marked in the right lower quadrant where there was marked spasticity.

The preoperative diagnosis was acute appendicitis.

Operation on October 7, 1939 was performed through a right lower Kammerer incision. The sigmoid was found drawn over to the right. The incision was closed and a left-sided incision made. Free feces was then found extruded from a "laceration" of the sigmoid. There was diffuse peritonitis. The perforation was closed by suture and the sigmoid area drained.

The final diagnosis was perforated diverticulitis with peritonitis.

Case 4: (Admission No. 503664) F. O., a 57-year-old man, was admitted to The Mount Sinai Hospital on May 31, 1928 with a 48-hour history of severe abdominal pain localizing in the right lower quadrant. He had had a preceding attack of pain which was thought at the time to be due to appendicitis.

On physical examination there was marked tenderness in the right lower quadrant. "Typical signs of peritonitis involving the lower half of the abdomen" were present.

The preoperative diagnosis was acute appendicitis.

Operation on June 1, 1928, was performed through a right rectus muscle splitting incision. There was a small amount of free peritoneal fluid. The sigmoid was over on the right side. It was thickened and "inflamed." The epiploicae were many times normal in size. The area about the sigmoid was drained. A normal appendix was removed.

The final diagnosis was acute diverticulitis.

COMMENT ON CASES 1, 2, 3, 4

In the four cases described above, the acutely inflamed sigmoid was found on the right side of the abdomen and was clearly responsible for symptoms and signs resembling acute appendicitis, by producing irritation of the parietal peritoneum in the right iliac fossa.

Case 5: (Admission No. 440483) A. N., a 71-year-old woman, was admitted to The Mount Sinai Hospital on May 16, 1939 with a one-day history of gradually increasing generalized abdominal cramps, associated with nausea. Her maximum pain was in the suprapubic region. She had a chill and temperature rise to 104.

On physical examination, temperature was 101. The abdomen was markedly distended. There was generalized tenderness, rebound, and spasm, all of which were most marked suprapubically and in the right lower quadrant. On rectovaginal examination, most tenderness was on the right.

The preoperative diagnosis was appendicitis with peritonitis.

Operation on May 16, 1939 was performed through a right lower rectus muscle-splitting incision. Thick, non-odorous pus was found coming from the pelvis. The appendix was seen to be normal. The sigmoid was easily delivered and found to be acutely inflamed and swollen. The epiploicae were thick and reddened. No perforation was seen. Appendectomy was performed. The sigmoid region was drained. The pathological report was acute peri-appendicitis. The culture of pus showed B coli.

The final diagnosis was acute perforative sigmoiditis.

COMMENT ON CASE 5

The signs in the midline and on the right side resembled those of pelvic appendicitis. The causative mechanism of the right-sided signs is not definitely known here, but from the operative description the easy delivery of the sigmoid through a right-sided incision suggests a position of the sigmoid to the right of the midline. The participation of the cecum in the general distention may have contributed to the right-sided signs.

PERFORATION WITH SPILLAGE OF FLUID TOWARD THE RIGHT

Case 6: (Admission No. 502490) M. F., a 57-year-old man, was admitted on March 6, 1943. For two weeks the patient had had mild lower abdominal cramps, more severe in the 24 hours before admission. Low grade fever was present for one day. Because no urine had been passed for 24 hours, catheterization was performed, with the evacuation of 100 cc. of bloody urine.

On physical examination the temperature was 101.6. The patient appeared very ill. There was distention and spasticity in the lower abdomen, slight bilateral tenderness, and distinct right costovertebral tenderness.

He was first admitted to the Urological Service. Cystoscopy and intravenous pyelogram on admission however were essentially normal. The abdominal signs became more localized on the right side where definite rigidity developed.

The diagnosis was altered to peritonitis due to ruptured appendix or cecal neoplasm.

Operation on March 7, 1946 was performed through a right lower rectus muscle-splitting incision. A great deal of cloudy fluid escaped from the peritoneal cavity. The cecum and appendix were normal. The gallbladder and stomach were normal on palpation. The small intestinal loops were packed away in order to visualize the sigmoid. A firm mass was seen and felt in the sigmoid on the right wall of which was a free perforation about 2 millimeters in diameter. The sigmoid was then exteriorized through a small left-sided incision and later excised.

The pathologic diagnosis was adenocarcinoma of the sigmoid with perforation.

Case 7: (Admission No. 375348) R. S., a 28-year-old man, was admitted on January 5, 1935. Seven months before admission the patient had sharp right lower quadrant pain lasting a few hours. Six weeks before admission he had dull pain in the right lower quadrant associated with vomiting and diarrhea. There was one shaking chill. Pain subsided after three days.

Fourteen hours before admission the patient was seized with periumbilical pain radiating to the right lower quadrant and to both testes, associated with nausea. He vomited once. He had one marked shaking chill. One soft stool was passed shortly after the onet of pain.

On physical examination the temperature was 103.4. There was tenderness, rigidity, and rebound tenderness in both the right and left lower quadrants. On rectal examination there was a bulge felt anteriorly above the prostate. There was marked tenderness on the right and less tenderness on the left.

The preoperative diagnosis was acute appendicitis with perforation.

Operation on January 5, 1935 was performed through a right lower rectus muscle-splitting incision. On opening the peritoneal cavity a thin, greenish-yellow fluid exuded. (Smear showed gram positive cocci.) The appendix was normal. Pus was then seen oozing from the pelvis. The loops of small intestine were packed away to expose the sigmoid. Close to the mesenteric border was a necrotic looking area, about 1.5 cms. in diameter, with a tiny hole in its center. The perforated area was sutured and drained.

The final diagnosis was inflammatory perforation of the sigmoid, probably due to diverticulitis.

Case 8: (Admission No. 394043) H. L., a 5-year-old boy, was admitted on June 3, 1936. He had a rectal polypectomy performed by suture and excision. A few bloody stools followed. Five days later he was seized with severe abdominal pain and a strong urge to defecate. He vomited once.

On physical examination, five hours after onset of symptoms, the patient had generalized abdominal tenderness and rigidity, definitely most marked in the right lower quadrant. Liver dullness was obliterated. (Roentgen examination of the abdomen showed air under both leaves of the diaphragm.) On rectal examination a small nubbin could be felt 1½ inches above the anus at the site of the polyp excision.

The preoperative diagnosis was ruptured viscus (possibly related to the rectal operation) or acute appendicitis with perforation.

Operation on June 3, 1936 was performed through a right paraumbilical muscle-splitting incision. When the peritoneum was opened, gas and odorless purulent fluid escaped. The cecum was delivered to expose a long, apparently acutely inflamed appendix, which was removed. Purulent fluid, however, kept coming from the rest of the peritoneal cavity. The stomach and gallbladder were palpated but no lesion was discovered. The sigmoid was then exposed. A firm, indurated mass was found about 6 centimeters above the level of the peritoneal reflection. Exudate overlay a perforation about one-half centimeter in diameter. The hole was closed and the area drained. The pathological report was periappendicitis.

The operator stated that he thought the perforation was too high in the sigmoid to be due to the polyp excision in the rectum.

COMMENT ON CASES 6, 7, AND 8

In these three cases the right-sided signs were apparently chiefly due to perforation on the right wall of the sigmoid with spillage of fluid toward the right. The absence of surrounding omentum may have more easily allowed this tracking of fluid.

In Case No. 8, the slightly reddened appendix was merely part of the contiguous inflammation as is seen by the pathological report. Further exploration before removal of the appendix would have been better, for the degree of inflammation in the appendiceal wall was clearly not sufficient to account for the clinical findings or the free pus in the peritoneal cavity.

Case 9: (Admission No. 420291) P. F., a 32-year-old woman, was admitted on February 15, 1938, because of sudden, severe, generalized abdominal pain beginning several hours before admission, followed by nausea and vomiting. The temperature rose abruptly to 103.8.

On physical examination the abdomen was distended. There was general abdominal tenderness, most marked in the right lower quadrant, where spasm was also present.

Operation was performed shortly after admission. A low midline incision was used. A large amount of free pus escaped from the peritoneal cavity. The sigmoid was found to be the site of a surrounding purulent infection. The appendix was removed. The pelvis was drained.

The lesion was finally discovered at autopsy to be due to an adenocarcinoma of the sigmoid with perforation into the cul de sac.

COMMENT ON CASE 9

Although most of the abdominal signs were clearly on the right, the sudden onset and rapid progression of generalized abdominal symptoms apparently suggested the possibility of a perforated left-sided lesion, for a midline incision was used. Cecal distention of course may well have contributed to the tenderness in the right lower quadrant.

Case 10: (Admission No. 465843) M. F., a 68-year-old woman, was admitted on November 27, 1940. She had had a sense of fulness in the lower abdomen for one day. A few hours later she was awakened from sleep by sharp, generalized abdominal pain. Her family physician found marked tenderness and spasticity limited to the entire right side of the abdomen. The pain persisted for several hours, was associated with nausea and vomiting, and finally seemed to localize in the right lower quadrant.

On physical examination the temperature was 101. Tenderness was found to be most marked in the left lower quadrant, although shortly before admission the signs were all right sided. Most of the tenderness on pelvic examination was on the right.

Operation on November 28, 1940 was performed through a right lower rectus muscle-splitting incision. No free fluid was found. The appendix was normal. A mass was found in the left lower abdomen involving the lower sigmoid and "adherent to all the surrounding structures." The mass had perforated into the general abdominal cavity. The sigmoidal area was drained.

Autopsy showed multiple diverticula with perforations, and a pericolonic abscess perforating into the posterior cul de sac. There was also a hemorrhagic pulmonary infarct.

COMMENT ON CASE NO. 10

Unmistakable right-sided symptoms and signs were present before admission to the hospital. After admission, the abdominal signs were chiefly left-sided and the pelvic findings right-sided. Though perforated diverticulitis was considered, the most likely diagnosis was appendicitis, for a right-sided incision was used. This case is classified as probably due to spillage of fluid toward the right, but adherence of small bowel to the sigmoid area may have been a cause of the referral of pain to the right.

Case 11: (Admission No. 512894) A. A., a 42-year-old man, was admitted on July 27, 1943. Sixteen hours before admission the patient developed lower abdominal crampy pain. He took a teaspoonful of Epsom salts. One hour later he began to have small bowel movements. Pain increased in severity and he vomited. Pain was predominantly in the midline, but also in both lower quadrants. He had a shaking chill.

On physical examination the temperature was 100.6. The abdomen was markedly distended. There was marked tenderness and spasm of the entire lower abdomen. On rectal examination, tenderness was limited to the right side.

The preoperative diagnosis was perforated appendicitis.

Operation on July 27, 1943 was performed through a right Kammerer incision. Thin cloudy fluid escaped from the peritoneal cavity. The cecum and appendix were normal. A mass was palpated and seen in the mid-sigmoid containing a perforation oozing fecal fluid. There was very little peritoneal reaction. The sigmoid was exteriorized through a left-sided incision and the involved area ablated. The pathologic report revealed a perforated diverticulitis.

COMMENT ON CASE NO. 11

The abdominal symptoms and signs were in the midline and in both lower quadrants, though rectal tenderness was on the right. The paucity of local reaction to the perforation, allowing the tracking of fluid throughout the peritoneal cavity, may have accounted for equal distribution of abdominal signs between right and left sides. Although the right-sided signs were not predominant (except for rectal findings) this case is included because to all observers the clinical picture simulated perforated appendicitis.

PERISIGMOIDAL ABSCESS WITH EXTENSION TO THE RIGHT

Case 12: (Admission No. 509520) Y. K., a 42-year-old woman, had had enorexia, cramps and diarrhea for three months, with pain in both lower quadrants. Five weeks before admission she was operated upon at another hospital through a right rectus incision and an abscess in the right iliac fossa was drained. The appendix, normal on pathological examination, was removed. Two weeks later the patient was admitted to The Mount Sinai Hospital after having a sudden chill, followed a few hours later by pain in the right lower quadrant.

On physical examination the temperature was 103.6. There was distinct tenderness in the right lower quadrant. A large, ill defined mass was palpable in the region of the right rectus incision scar. On pelvic and rectal examinations, tenderness was present on both sides.

Under observation the symptoms and signs progressed. The preoperative impression was that the right lower quadrant abscess had probably originally been due to a terminal ileitis with perforation.

Operation was performed through an incision lateral and parallel to the previous right rectus scar. A large omental mass extending towards the left side was found in the right iliac fossa. This area was drained though no pus was encountered. On the fifth postoperative day there was spontaneous discharge of copious foul-smelling pus from the drainage tract. The lesion was water found at another operation to be a rectosigmoidal carcinoma with perisigmoidal abscess.

Case 13: (Admission No. 418912) E. G., a 55-year-old woman, was admitted on January 11, 1938. For one year she had occasional blood streaked stools and a gradual loss in weight. Two weeks before admission the patient had a shaking chill followed by pain in the right lower quadrant which became constant.

On physical examination the abdomen was soft but distended. There was deep tenderness above Poupart's ligament on the right with some muscular spasm. A fulness could be felt in the right lower quadrant though no distinct mass was palpable. Rectal and pelvic examinations disclosed a tender mass to the right of the cervix.

The admission diagnosis was appendiceal abscess. The possibility of a perisigmoidal abscess due to a perforating carcinoma was considered, but the symptoms and signs were so clearly on the right that a right-sided incision was made for exploration.

Operation on January 14, 1938 was performed through a low right rectus muscle-splitting incision. There was thin, free peritoneal fluid. An abscess was found in the

pelvis behind the uterus, extending up toward the cecum. After suitable walling off with pads, the abscess was drained.

At autopsy the rectosigmoid contained a carcinoma which had perforated into an abscess cavity lying behind the right tube and ovary.

Case 14: (Admission No. 418348) R. S., a 48-year-old woman, was admitted on December 29, 1937 with a five-week history of persistent lower abdominal cramps, marked constipation except for one episode of diarrhea, anorexia, loss of weight and fever. Barium enema at another hospital two weeks before admission was said to show no abnormalities.

On physical examination the temperature was 104. The abdomen was distended and slightly tender, chiefly in the right lower quadrant, where a sausage-shaped mass was palpable. On pelvic examination a firm, spherical, fixed mass was felt in the cul de sac. The pelvic mass seemed to be continuous with the abdominal mass.

The possible diagnoses considered included perforation of the small bowel, carcinoma of the sigmoid, and finally uterine fibroids with degeneration.

Operation on January 1, 1938 was performed through a left lower rectus incision. A huge mass was found filling the pelvis and extending up as far as the umbilicus. Across the front of the mass coursed the sigmoid. The mass was entered on the right side with evacuation of pus. It was then seen that the perforation of a sigmoidal tumor was the cause of the abscess.

The final diagnosis was carcinoma of the sigmoid with perisigmoidal abscess.

COMMENT ON CASES NOS. 12, 13, AND 14

Right-sided symptoms and signs in these three cases were caused by extension to the right of an abscess arising in the pelvis around the sigmoid and rectosigmoid.

In Case No. 12 the bulk of the abscess presented itself in the right iliac fossa. In Case No. 13, the diagnosis of perisigmoidal abscess due to carcinoma was considered but the virtual limitation of symptoms and signs to the right side of the abdomen prompted operation through a right rectus incision. In Case No. 14, although the abdominal signs were mainly on the right and the mass was felt to the right of the umbilicus, a left-sided incision was used because a degenerated uterine fibroid was thought to be the cause. However, at operation the sigmoid was found extending toward the right and the most accessible part of the abscess was also to the right of the midline.

Case 15: (Admission No. 513892) I. K., a 41-year-old woman, was admitted on December 4, 1943. One month before admission the patient was seized with sudden, severe, right lower quadrant pain requiring a hypodermic injection. The pain subsided but then recurred intermittently. It was always located in the right lower abdomen. The day before admission the pain increased in severity and was associated with nausea and vomiting.

On physical examination the temperature was 101.8. There was direct and rebound tenderness in the right lower abdomen. A midline mass was palpable above the pubis. On pelvic examination a firm, very tender mass, the size of a grapefruit, was felt overlying the uterus and filling the right adnexal region.

The preoperative diagnosis was right ovarian neoplasm with torsion.

Operation on December 4, 1943 was performed through a midline incision. On separating the omentum, which was adherent to the pelvic structures, an abscess cavity was entered to the right of the uterus, yielding fecal smelling pus. The sigmoid loop lay across the midline and was adherent to the uterus. The actual point of perforation was

not seen, but one wall of the abscess cavity was composed of an indurated area in the sigmoid. The sigmoid was exteriorized and its limbs were approximated to form a spur.

The final diagnosis was perisigmoidal abscess.

COMMENT ON CASE NO. 15

The symptoms and signs were apparently due both to the course of the sigmoid across the midline and to the location of the abscess to the right of the uterus.

Case 16: (Admission No. 513839) M. B., a 65-year-old woman, was admitted on December 2, 1943. After being previously well, the patient had sudden right lower quadrant pain associated with nausea and vomiting one day before admission. The symptoms gradually increased in severity.

On physical examination the abdomen was distended. Marked tenderness and spasm were present in the right lower quadrant. There was also slight tenderness in the left lower quadrant.

The preoperative diagnosis was acute appendicitis with possible perforation.

Operation on December 2, 1943 was performed through a right lower rectus muscle-splitting incision. There was a small amount of free peritoneal fluid. The appendix was normal. The sigmoid was exposed. An area of induration was found in the sigmoid with a surrounding abscess which was drained.

The final diagnosis was perisigmoidal abscess due to perforated diverticulitis.

COMMENT ON CASE NO. 16

Typical symptoms and signs of acute appendicitis were caused by a perisigmoidal abscess, but the mechanism of the crossed symptomatology in this case is not clear from the operative description. Here, too, cecal distention may well have been partially responsible for the symptoms and signs on the right.

ADHERENCE OF RIGHT SIDED STRUCTURES TO THE SIGMOID

Case 17: (Admission No. 415178) F. C., a 39-year-old man, was admitted on October 9, 1937. For eight days before admission the patient had lower abdominal cramping pain. There was mild nausea and one episode of vomiting. His physician diagnosed "subsiding appendicitis." On the night before admission, following an enema, the lower abdominal cramps became more severe and finally shifted to the right lower quadrant.

On physical examination the temperature was 102.8. Tenderness and rigidity were found in both lower quadrants, most marked on the right. There was a firm, tender mass filling most of the right lower abdomen and extending into the left lower quadrant. On rectal examination, tenderness was felt on the right.

The preoperative diagnosis was appendiceal abscess.

Operation on November 6, 1937 was performed through a right lower rectus muscle-splitting incision. The appendix was found lying free, "moderately injected," with an apparently blunted tip. Appendectomy was performed. Further exploration, however, revealed an abscess in the left iliac fossa, surrounded by omentum and loops of small intestine. The cavity was entered and drained. Later roentgen studies of the sigmoid showed diverticulitis.

COMMENT ON CASE NO. 17

Although the sigmoid and the perisigmoidal abscess were located in the left iliac fossa, the symptoms and presenting mass on clinical examination were

on the right. The symptoms may have been due to the adherence of the small bowel loops to the perisigmoidal abscess. The mass palpated on the right side was apparently matted small intestinal loops forming and covering the abscess wall. It is of interest to note that right-sided symptoms again were the presenting complaints one and a half years after recovery from the first attack.

The following case was previously described by Arnheim[2] in his study of diverticulitis, but is here presented as another instance of right-sided symptoms produced by adherence of right-sided structures to a sigmoid lesion.

Case 17A: (Admission No. 345651) P. K., a 57-year-old man, was admitted on November 14, 1932 with the history of right lower quadrant pain and constipation for four months, loss of weight for two months, and a chill two weeks before admission.

On physical examination the temperature was 98.6 degrees. Tenderness was present in the right upper abdomen. Barium enema studies, however, revealed many diverticula of the sigmoid with spasm.

Operation was performed through a left-sided incision. The sigmoid was short and fixed, and contained several diverticula. An abscess posterior to the sigmoid was entered and 2 ounces of thick, creamy, odorless pus was evacuated.

At autopsy there was an inflammatory mass involving the sigmoid, omentum, and a loop of ileum. At the site of attachment of the ileum to the sigmoid there was an ileosigmoidal fistula.

SITUS INVERSUS

Case 18: (Admission No. 527537) F. K., a 63-year-old woman, was admitted on November 18, 1944. In the three years before admission the patient had attacks of right lower quadrant pain associated with the urge to defecate, relieved by passage of gas. She had lost 15 pounds in the past year. Four days before admission a particularly severe attack of right lower quadrant pain occurred, associated with left flank pain and nausea. A mild chill occurred.

On physical examination the temperature was 100.4. The abdomen was soft. In the right lower abdomen extending out to the right flank was an ill defined, soft, tender mass which did not move on respiration. There was no tenderness on pelvic and rectal examinations.

The preoperative diagnosis was appendiceal abscess. The possibility of a cecal neoplasm or cecal diverticulitis was considered.

Operation on November 20, 1944 was performed through a right lower rectus incision. On opening the peritoneum, the sigmoid presented itself in the wound. A mass was present in the right wall of the sigmoid which seemed to be bound to the right lateral peritoneum. The area around the mass was drained and the wound closed, leaving the subcutaneous tissues packed open.

Barium enema disclosed a situs inversus of the intestinal tract, and showed diverticula with spasm in the sigmoid colon.

COMMENT ON CASE NO. 18

The sigmoid is a right-sided organ in situs inversus and therefore would be expected to produce symptoms on the same side. This case is included merely for completeness to illustrate one of the possible mechanisms of the production of right-sided symptoms by a sigmoid lesion.

SUMMARY AND CONCLUSIONS

1. Right-sided abdominal symptoms and signs are often produced by sigmoidal lesions. Acute appendicitis is the commonest preoperative diagnosis in these instances.

2. The principal causes for the right-sided symptomatology in acute diverticulitis and carcinoma of the sigmoid are:

a. Sigmoid lies on the right side of the abdomen as a result of its mobility or the anatomical variations in its course;

b. Perforation of the right wall of the sigmoid with spillage of exudate into the right iliac fossa;

c. Extension of a perisigmoidal abscess to the right;

d. Adherence of right-sided structures to the sigmoidal lesion;

e. Marked distention of the cecum;

f. Situs inversus.

Eighteen cases are reported in illustration (5 with carcinoma and 13 with diverticulitis).

3. Inspection of the sigmoid should be included in the exploration of the abdomen before removal of the appendix, when at operation insufficient cause for the clinical findings is discovered in the right iliac fossa.

The cases in this paper are from the Surgical Services of Drs. John H. Garlock, Ralph Colp, and Harold Neuhof, and include three private cases from the services of Drs. Albert A. Berg, Leon Ginzburg, and Myron Sallick.

BIBLIOGRAPHY

[1] Allen, P. D.: Peritonitis Secondary to Perforation in Carcinoma of the Colon. New York State J. of M., **43**: 1732, 1943.

[2] Arnheim, E. E.: Diverticulitis of the Colon with Special Reference to the Surgical Complications. Ann. Surg., **112**: 352, 1940.

[3] Young, E. L., and E. L. Young: Diverticulitis of the Colon, New England J. of Med., **230**: 33, 1944.

[4] Cave H., and W. E. Alsop: Diverticulitis of the Colon. Surg. Clin. of N. A., 390, 1946.

[5] Morton, J. J.: Diverticulitis of the Colon. Ann. Surg., **124**: 725, 1946.

[6] Rankin, F. W., and A. E. Grimes: The Surgical Treatment of Diverticulitis. South. Surgeon, **1**: 7, 1938.

[7] Jones, T. E.: Diverticulitis and Diverticulosis. Surg. Clin. of N. A., 1105, 1939.

[8] Beer, E.: Some Pathological and Clinical Aspects of Acquired Diverticula of the Intestine. Am. J. M. Sc., **137**: 135–145, 1904.

[9] Erdmann, J. F.: Acute Diverticulitis of the Colon. Am. J. Obst. and Gynec., **11**: 609, 1926.

[10] Abell, I.: Diagnosis and Treatment of Diverticulitis and Diverticulosis. Surg., Gynec. & Obst., **60**: 370, 1935.

[11] Ransohoff, N.: Perforative Sigmoiditis in Children. Ann. Surg., **58**: 218, 1913.

[12] Rankin, F. W.: Surgery of the Colon. New York, D. Appleton & Co., 1926.

[13] Ochsner, A.: Surgery of the Intestine. Nelson Loose Leaf of Surgery, 1932.

[14] Babcock, W. W.: Textbook of Surgery. Philadelphia, W. B. Saunders Co., 1935.

[15] Christopher, F.: Textbook of Surgery. Philadelphia, W. B. Saunders Co., 1942.

[16] Homans, J.: Textbook of Surgery. Springfield, Charles C. Thomas.

17 Bearse, C.: Acute Perforative Diverticulitis of the Colon in Young Persons. J. A M. A., **113**: 1720, 1939.

18 Brown, P. W.: Treatment and Prognosis of Diverticulitis of Colon. Am. J. Surg., **46**: 162, 1939.

19 Rankin, F. W., and P. W. Brown: Diverticulitis of the Colon. Surg. Gynec. and Obst., **50**: 836, 1930.

20 Dixon, C.: Diverticulitis. Am. J. Surg., **46**: 600, 1939.

21 Strauss, A. A., S. F. Strauss, and H. A. Strauss: Differential Diagnoses of Diseases of the Colon. Surg. Clin. of N. A., **203**, 1939.

22 Rankin, F. W., J. A. Bargen, and L. A. Buie: The Colon, Rectum and Anus. Philadelphia. Saunders, 1935.

23 Lynch, J. M.: Diverticula and Diverticulitis. J. A. M. A., **98**: 973, 1932.

24 Graham, R.: Diverticulitis of the Sigmoid Colon. Canad. M. A. J., **36**: 1, 1937.

25 Laufman, H.: Collective Review. Internat. Abstr. Surg., **73**: 722, 1941.

26 Ssoson-Jaroschewitsch: Die Analyse der Variationen des S romanum. Arch. f. Klin. Chir., **125**: 283, 1923.

27 Colp, R.: Colonic Spasm as the Cause of Intestinal Obstruction. Surgery, **10**: 270, 1941.

28 Kantor, J. L.: Perforating Ulcers of the Cecum. J. A. M. A., **79**: 59, 1922.
Dickinson, G. K.: Perforating Ulcers of the Cecum. J. A. M. A., **79**: 1792, 1922.

29 Kantor, J. L.: Common Clinical Types of Appendicitis. Med. Clinics of North America, **5**: 1081, 1922.

NEW DONOR AREAS IN SKIN GRAFTING

DONALD EARL BARKER, M.D.
PHILADELPHIA, PA.

WITH THE INTRODUCTION of the dermatome in 1939,[1] the taking of calibrated skin grafts became fairly simple. However, it soon became apparent that this was limited in its scope by the irregularities in the surface of the prospective donor areas. Adequate grafts could be taken from the thighs, abdomen and back in a thin individual, and in addition from the chest in an obese individual. Many cases, however, are seen in which the above donor areas have been destroyed by burn. The only areas of normal skin remaining are on the chest, upper back, and scapula where by the time the individual is ready for grafting, the surface is too irregular to take a graft because of the

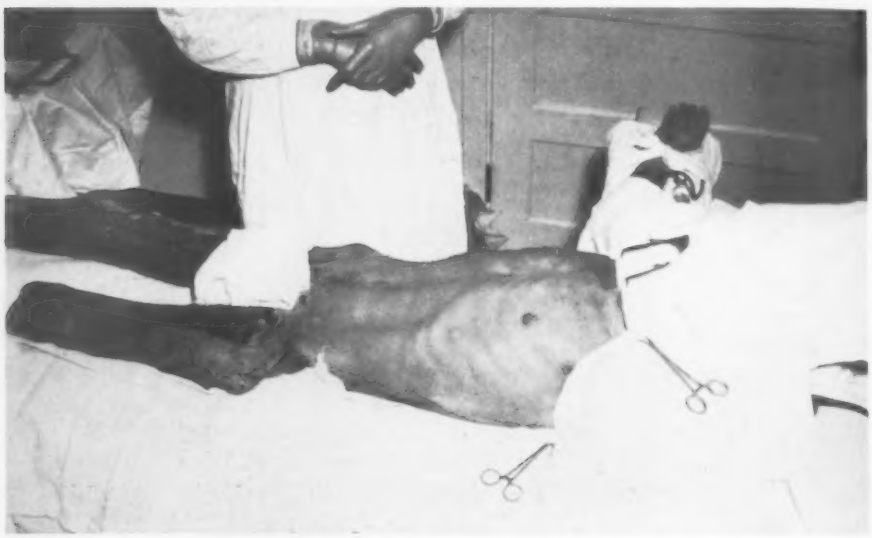


FIG. 1.—Chest showing prominent ribs.

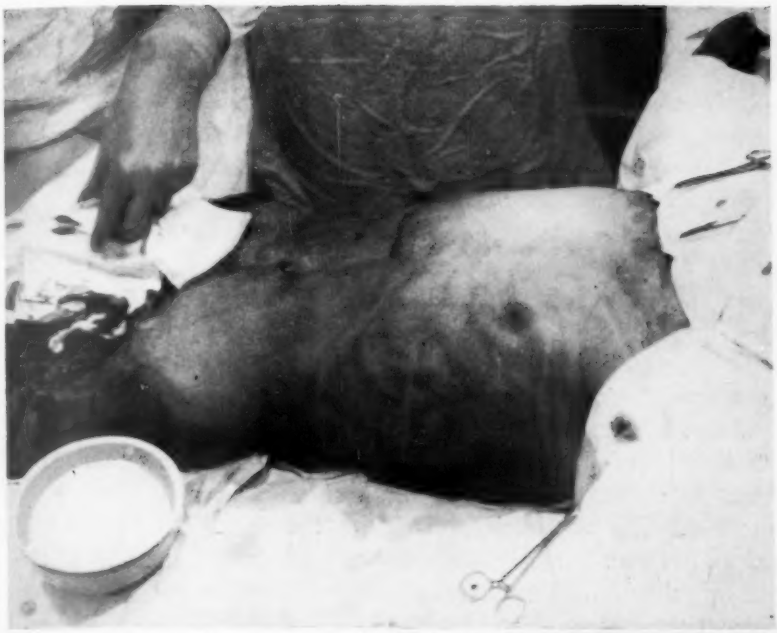
wasting of the subcutaneous tissue and the protuberance of the underlying bone. The taking of Thiersch grafts with the Ferris-Smith knife is usually limited to the thighs or the upper arms.

Grafts can be taken from practically all irregular areas by injecting saline beneath the skin, with obliteration of the depressions. A search of the literature failed to reveal any article in which this had been described. This method may possibly have been used elsewhere unknown to the present author; however it has apparently not been published in medical literature so that the other surgeons might utilize it.

TECHNIC

An area of normal skin slightly larger than the dermatome is marked out with methylene blue. Using a 30 cc. syringe and a long needle, normal saline

FIG. 2



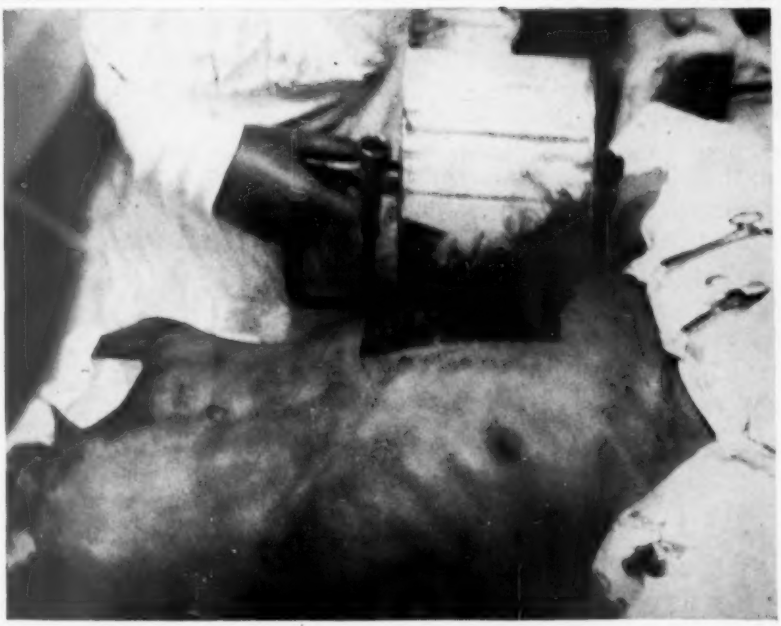


FIG. 3

FIG. 2.—Smooth contour after subcutaneous saline.
FIG. 3.—Graft taken from area.

solution is injected beneath the skin in the area outlined by the methylene blue. Care should be taken that the needle is not put through the skin at any point in the rectangular area outlined by the ink. In the first case in which this was used the needle entered the normal skin area where the graft was to be taken, and the saline leaked out onto the donor skin before the skin cement was applied. After all of the irregular areas have been obliterated the area is cleansed well with ether and the Padgett cement is applied. After the cement has dried thoroughly for five minutes the graft is taken in the usual manner.

The first case upon which this was used was an emaciated male, age 22, burned in an airplane crash. The only normal skin available was over the sternum and the scapula. Grafts were taken from both of these areas in this patient. In another patient, although some other donor areas were available it was felt that the work could be completed faster by using sheets of skin larger than those available with the Padgett dermatome. The dermatome used here was a special one taking a graft 6 by 10 inches, or nearly twice the area of skin taken with the Padgett machine. The use of this type of dermatome is usually limited to circular grafts of the thigh, or the occasional graft from the chest in an obese individual. Figure 1 shows a preoperative view of the donor area, showing marked irregularity of the chest. This patient had an estimated 40 per cent burn on admission. Figure 2 is a close-up of the area demonstrating the marked smoothness of the donor skin that can be obtained with subcutaneous saline. Figure 3 shows a graft of 60 square inches taken from the chest. This method can also be used for taking free hand grafts. An irregular chest, scapular, or back area can be used after subcutaneous injection of saline.

SUMMARY

A method is presented whereby irregular skin areas may be utilized as donor areas using the dermatome, or by cutting the graft free hand. This is accomplished by injection of normal saline beneath the prospective donor area.

REFERENCES

Padgett, Earl C.: "Calibrated Skin Grafts." Surg., Gynec. and Obst., 69: 779–793, 1939.

GASTRODUODENAL ULCER, A SPASTIC DISEASE

I. Boerema, M.D.

Professor of Surgery, University of Amsterdam, Holland

Although hundreds of gastrectomies are performed daily there is still no unanimity of opinion as to the underlying cause of ulcers of the stomach and duodenum. Clinical, pathologic and experimental investigations have failed to solve the problem completely. The German occupation of Holland, 1940–1945, during which the entire population of nine million people were medically, *de facto* experimental subjects,* furnished a great deal of material which may throw some light on the theory of ulcer formation.

Both psychical and alimentary disturbances during the occupation may have affected the gastrointestinal tract. In Holland, as in the United States, an increased number of ulcers occurred immediately after the outbreak of war. This early rise may be attributed to psychic factors such as fear, sorrow, and rage, since at that time there was no appreciable change in food supplies. Shortly thereafter important changes in diet of the Dutch people occurred, and this was accompanied by a great increase in incidence of gastroduodenal ulcers. For many years the daily intake consisted of a total of about 1600 calories, with a relative diminution in consumption of protein and of fat, and an increase in carbohydrates. Potatoes, a food high in cellulose, were eaten in much larger quantities than normal. These changes produced an increased peristalsis throughout the gastrointestinal tract of nearly the whole population. Many persons even suffered from non-bacterial diarrhea throughout the entire period of the occupation.

The incidence of other diseases was also changed during the occupation. Some increase in the number of operations for gallstones was noted. Lack of fat probably kept this increase small, for immediately following the liberation, when the fat supply improved, we saw innumerable gallbladder cases. Volvulus, especially of the sigmoid, strangulated herniae and anal fissures were observed in greater numbers, while there was a diminution in the incidence of acute appendicitis (Table I).*† Volvulus and anal fissure are conditions known to be associated with either increased peristalsis or spasticity of part of the intestinal tract. Spasticity of the sphincter of the anus can be directly observed in cases of anal fissure. Increased peristalsis, preventing stagnation of secretions in the appendix, may also explain the decrease in appendicitis.

* The observations of this huge experiment were collected and studied by a team of 21 investigators, most of them connected with Dutch Universities, and described in "Medical Experiences during the Occupation of The Netherlands 1940–1945."

† Figures in this paper do not include the years 1944 and the first half of 1945. During this time the city populations were so changed because of inundations, displacements and bombardments that a quantitative comparison with normal times is impossible.

These observations lead to the conclusion that there was an increase in peristalsis and a tendency to spasm in the intestinal tract of many Dutch people during the occupation. It seemed probable that similar conditions might also exist in the stomach and duodenum. Table II gives the statistics on ulcer cases operated upon during this period. It is our theory that the increase in the number of ulcers may be a direct result of the spastic condition assumed to exist also in the upper intestinal tract.

TABLE I.—*Incidence of Volvulus, Anal Fissure and Appendicitis*

	1938	1939	1940	1941	1942	1943	1944
Operated cases of volvulus, large and small intestine.. (All clinics in Groningen)	1	4	5	6	15	11	..
Cases of anal fissure (Surgical clinic, Groningen)							
Men	2	5	3	9	6	8	13
Women	2	5	0	8	6	11	9
Total	4	10	3	17	12	19	22
Cases operated appendicitis (all hospitals in Groningen)							
Men	481	468	345	346	343	318	...
Women	464	494	375	398	400	379	...
Total	945	962	720	744	743	697	...

TABLE II.—*Increase in Frequency of Gastroduodenal Ulcer*

	1938	1939	1940	1941	1942	1943
Operated cases non perforated gastric and duodenal ulcers (All hospitals, Groningen).						
Men	81	96	86	144	158	260
Women	10	21	22	28	21	25
Total	91	117	108	172	179	285
Operated cases gastric ulcers (University Hospital, Groningen)						
Men	2	11	14	36	15	27
Women	2	1	1	4	1	0
Total	4	12	15	40	16	27
Operated cases duodenal ulcers (University Hospital, Groningen)						
Men	18	14	15	33	16	35
Women	1	2	0	4	0	0
Total	19	16	15	37	16	35

Pyloric hypertrophy in adults in Holland was frequently observed, an indication of increased frequency of a spastic condition of the stomach. Roentgenologists demonstrated many cases where the entire antrum of the stomach was rigid: laparotomy revealed neither cancer nor ulcer, but hypertrophy of the pylorus. Spasm of the entire antrum probably occurred in such cases. One such patient, a woman of 72, had heavy cramps in the stomach *and diarrhea,* after meals. Cancer was suspected because of a stiff antrum, but resection of part of the stomach and of the upper part of the duodenum

showed only a very thick pylorus. Such direct observations of increase of frequency in spasm in the stomach sustain the neurogenic theory of ulcer of Von Bergmann. According to this theory, local spasms cause ischemia in the wall of the gastroduodenum, after which digestion takes place of it.

Acidity of the gastric secretion decreased, rather than increased, during the occupation. Hence these observations do not support the theory of high acidity as the cause of gastroduodenal ulcer. Neither can the increase in ulcer cases be attributed to emboli in the gastroduodenal wall. Actually, as indicated in Table III, thrombo-embolism decreased in frequency, and even almost disappeared during the last two years before the liberation, but resumed its prewar incidence within three months thereafter. A fourth theory, that gastroduodenal ulcer is a complication of chronic gastritis, is not borne out by our observations. Although there was a great increase in diarrhea among the Dutch population, this was mostly nonbacterial in origin. Studies of infection in operative wounds and of hematogenous osteomyelitis indicate that the people of Holland were no more susceptible to infection during the first three years of the occupation than before the war.

TABLE III.—*Incidence of Postoperative Thrombosis and Embolism*

	1938	1939	1940	1941	1942	1943
Postoperative thrombosis and embolism (Surgical Clinic, Groningen)	30	33	28	12	13	14
Fatal pulmonary embolism	11	17	7	3	2	1
Non fatal pulmonary embolism (Postmortem examinations, Binnen Gasthuis, Amsterdam)	61	76	54	20	14	9

TABLE IV.—*Incidence of Prolapse of Rectum and Anus*

	1938	1939	1940	1941	1942	1943
Operated prolapsis ani and recti (Surgical Clinic, University Hospital, Groningen)	4	12	16	18	28	24

Our conclusion is, therefore, that gastroduodenal ulcer is a spastic disease whose etiology is best explained by the theory of Von Bergmann.

A study of spastic conditions in children of various ages was made in order to determine whether psychic or nutritional factors were more important in causing intestinal spasm. Breast-fed babies may be assumed to be relatively uninfluenced by war, either psychically or in actual feeding. In this group we observed no changes during the occupation from pre-war levels of frequency of pylorospasm. Children of one year were at an age to be somewhat affected by food changes. This group showed a small but distinct increase in the incidence of ileocecal intussusception, a spastic disease occurring most frequently at this age. Children of four to five years, still uninflu-

enced psychically by the war, suffered as much as adults from the change in food supply. In this age group, as shown in Table IV, a substantial increase in prolapse of the mucous membrane of the rectum and anus occurred. This condition is associated with an increase in peristalsis and spasm of the lower part of the bowel. These observations of children indicate that changes in the physiology and pathology of the bowel are influenced in the first place by the food ingested.

The above considerations cause us to examine the rationale for the hundreds of resections for treatment of gastroduodenal ulcer performed daily. Our experiences during the occupation lead us to conclude that diminishing the gastric acidity can not be the cause of the success of gastric resection. Removal of the ulcer, *per se,* is not the reason, as from our experience it is immaterial whether the duodenal ulcer is removed or left *in situ,* provided at least half the stomach, including the pylorus, is removed. The chief effect of resection seems to us to be the removal of that part of the gastroduodenum most susceptible to the spasm which we assume to be the direct cause of the ulcers.

This study, while not affecting therapy, has, in fact, led us to the conviction that gastroduodenal ulcer is a spastic disease.

RATIONALE OF PARENTERAL GLUCOSE FEEDING IN THE POSTOPERATIVE STATE

MORTON D. PAREIRA, M.D., AND MICHAEL SOMOGYI, Ph. D.

FROM THE DEPARTMENT OF SURGERY AND LABORATORIES,
JEWISH HOSPITAL OF ST. LOUIS, ST. LOUIS, MISSOURI

GLUCOSE serves two purposes in the postoperative state if administered properly: It acts to prevent starvation ketosis, and to minimize protein catabolism by exertion of its protein sparing effect.

Ketosis always occurs when the liver lacks adequate glycogen stores. Muscles and other tissues may be provided with all the carbohydrate necessary for their normal metabolism, but ketosis still develops if the liver is deprived of available glycogen supply. Ketosis, if it emerges and is allowed to persist, leads to a progressive deterioration of the architecture of the hepatic cell, since the ketone bodies, *i.e.*, β-hydroxybutyric and acetoacetic acids, which are formed exclusively in the glycogen deprived liver, alter the hydrogen ion concentration, and, at the same time, the electrolyte pattern of the cells. These changes inevitably interfere with the enzymatic reactions and the immunologic functions of the liver. In a word, normal hepatic function is incompatible with ketosis.

Lack of hepatic glycogen initiates, in addition to ketosis, a second adverse process, in that it compels the body to burn increased amounts of protein as well as fat. It is well known that depletion of the protein reserves of the liver is as harmful to normal liver function as exhaustion of the glycogen stores, both being essential elements in the normal physico-chemical structure of the liver cell.

In the immediate postoperative period one or more of three factors may be conducive to hepatic glycogen deficiency. One factor is the glycogenolytic effect of certain anesthetics which accelerates the conversion of liver glycogen into blood sugar and thereby substantially depletes the glycogen reserve. A second factor is the often necessarily imposed inadequacy of oral food intake directly following operation which, especially when superimposed upon either of the other factors, leads to rapid exhaustion of the hepatic glycogen content. A third factor may be the surgical disease itself, especially if it be one with which inanition, hepatitis or circulating hepatotoxins are associated.

As preventive measures against the injurious effects of surgical anesthesia, proper preoperative dietary measures have been suggested during the past four decades by many experimental investigations and clinical observations. The studies of H. G. Wells,[1] Opie and Alford,[2] and E. A. Graham,[3] to quote but a few representative examples, emphasized the importance of building rich hepatic glycogen reserves as a defense against liver injury by anesthetics;

This work was carried out with the aid of a grant from the Louis M. Monheimer Memorial Fund.

Whipple and his collaborators[4, 5] demonstrated the efficacious protective effect of diets which are rich in protein and low in fat content.

Appropriate preoperative dietary measures, in addition to protecting the liver against injury by anesthesia, help to counteract the ill effects of postoperative difficulties in the feeding of the patient. But regardless of how well the liver had been stocked with proteins and glycogen, the reserves are rapidly exhausted; hence it is necessary to resort to parenteral glucose feeding as soon as possible following operation in order to prevent hunger ketosis. This parenteral alimentation is necessarily linked with supplying the necessary amounts of water and electrolyte. The fluid and electrolyte requirements of the postoperative patient have been quantitatively elaborated by many workers. The same cannot be said in regard to parenteral glucose therapy. There are divergencies of opinion and practice, stemming in part from lack of appreciation of the quantitative factors involved, and in part from disregard of certain, pertinent physiologic laws. The studies reported here are concerned with the application in practice of these factors.

The amount of glucose needed in the postoperative period is decided by the purposes of its administration. Its primary purpose is to provide the liver with enough glycogen for the prevention of ketosis. Our studies have shown that on an average of 200 Gm. of glucose per 24 hours will serve this purpose providing that it is well spaced throughout the period. There are instances in which 150 Gm. of glucose will suffice; these are cases without appreciable liver damage and with adequate preoperative preparation. Again, there are instances when 200 Gm. of glucose per 24 hours is insufficient to forestall ketosis; these are the cases in poor nutritional state at the time of operation due either to inanition or to seriously impaired liver function. While the administration of 150 to 200 Gm. of glucose a day may suffice to prevent ketosis it will not supply the total energy requirements of the body; as a consequence undue quantities of protein will be consumed for fuel. By the simple means of increasing the glucose supply one can spare appreciable amounts of protein from being wasted in this manner with the additional advantage of preventing the inevitable formation of ketone bodies in the course of protein catabolism. Therefore, simply as a means of good physiologic economy, it is desirable to supply from 200 to 350 Gm. of glucose per 24 hours as a general rule. The upper limit applies to cases with known or suspected liver damage and to cases in which a poor nutritional state could not be remedied prior to operation.

The concentration of glucose solution to be employed is rather clearly defined by the quantitative requirements. To meet and yet not to exceed the postoperative fluid needs the necessary amount of glucose can be given only in solutions of not less than 10 per cent concentration. To meet the glucose requirement with less concentrated solutions would necessitate the administration of frequently excessive, and often prohibitive, amounts of fluid. It is a widespread view that 10 per cent glucose solution causes diuresis and thrombosis. Many, in holding this view, feel it mandatory to adhere to the use of

5 per cent solutions. Regardless of the validity of these objections, 5 per cent solutions often will not meet the physiologic needs. The amount of glucose being thus limited, ketonuria is in many instances virtually inevitable. An example to illustrate this fact is given in Table I.

The patient was a young woman in good health except for chronic cholecystitis with cholelithiasis, for which cholecystectomy was performed. There had been no episodes of biliary obstruction or clinical cholangitis. The patient, as indicated in Table I, was given 1500 cc. of 5 per cent glucose solution on the day of operation, and 2000 cc. daily during the ensuing two days. On the second postoperative day when oral feeding was attempted, the patient was nauseated sufficiently to preclude it. We suggested the possibility of ketosis, and a urine sample was found to show "three plus" acetone. We then suggested the administration of 2500 cc. of 10 per cent glucose solution.

TABLE I.—*Showing a Case in which Ketosis Was Not Prevented by 5 Per Cent Glucose Solution, But Was Promptly Controlled by 10 Per Cent Solution.*

Time After Start of Glucose Infusion Hours	Acetone (urine)	Amount of Glucose Infused
Nov. 10	*	1500 cc. 5% solution
Nov. 11	*	2000 cc. 5% solution
Nov. 12	+++	2000 cc. 5% solution
Nov. 13 (prior to infusion)	+++	
10:20 a.m.	+++	2500 cc. 10% solution started; infused over
11:30 a.m.	+	the course of the ensuing 7 hrs., beginning
12:30 p.m.	±	at rate of 250 cc./hr. and gradually acceler-
2:30 p.m.	0	ating to 500 cc./hr. during last 2 hrs.
3:30 p.m.	0	
Nov. 14		(Fed orally)

* Not tested.

Since it is well known that starvation ketosis entails a diabetic condition, that is to say, impaired carbohydrate tolerance, it was further suggested that the infusion be started at a rate of 250 cc. per hour and then gradually accelerated from hour to hour, until the rate should become 500 cc. per hour during the sixth and seventh hours. After the onset of this procedure ketonuria rapidly decreased to "one plus" during the first, and to a trace during the second hour of infusion, and completely disappeared during the third hour. The precaution as regards the initial low rate of infusion was taken in order to prevent undue loss of glucose in the urine. During the second and third hours of the infusion, when the rate was slightly increased, there was a loss of 8 Gm. of glucose in the urine. Thenceforth there was no glycosuria despite a further increase in the infusion rate. By the time the infusion was completed the patient was amenable to oral feeding, and remained so throughout the postoperative course. We believe that it is fair to infer from these facts that the nausea and anorexia were due to ketosis. This ketosis was quite evidently starvation ketosis, which 2000 cc. of 5 per cent glucose solution daily failed to avert.

It is well recognized that ketosis can be the cause of more serious consequences than nausea and anorexia. More severe forms of ketosis may lead to acidosis, with ensuing depletion and derangement of the electrolyte pattern of the tissue cells. Such changes frequently induce smooth muscle spasm, which often is reflected in acute abdominal pain.[6] An example of this phenomenon, from among our observations, is that of a patient following supravaginal hysterectomy. This patient was nourished postoperatively with amigen in 5 per cent glucose solution and small amounts of low caloric fluid by mouth. On the fifth postoperative day it came to our attention that the patient was beset by nausea, vomiting, and excruciating epigastric pain. Severe ketosis was suspected. A urine sample showed "four plus" acetone and "four plus" acetoacetic acid. Infusion of 2500 cc. of 10 per cent glucose solution was recommended. As in the preceding case, the existent ketosis was quite likely to

TABLE II.—*Showing that Infusion of 10 Per Cent Glucose Causes No Diuresis When Not Accompanied by Glycosuria.*

10% Glucose Infused in 2.5 to 3 hr. Period		Urine Excreted		
Time of Start	Volume cc.	Time	Volume cc.	Glycosuria Gm.
Nov. 9th				
3:40 a.m.	1000	From operation to 9 p.m.	175	0
		9 p.m. to 8:30 a.m.	450	0
Nov. 10th				
1:00 a.m.	1000	8:30 a.m. to 10 p.m.	480	0
8:00 a.m.	500			
	2500 cc. intake		1105 cc. output	
Nov. 14th				
4:00 p.m.	1000	7 a.m. to noon	145	0
11:30 p.m.	1000	Noon to morning	800	Trace
Nov. 15th				
3:00 p.m.	1000	7 a.m. to noon	100	Trace
		Noon to 5 p.m.	100	0
		5 p.m. to 7 a.m.	700	0
	3000 cc. intake		1845 cc. output	

have impaired carbohydrate metabolism, hence the glucose was administered over a period of six hours, starting at a slow rate which was then gradually increased. After the fourth hour of infusion ketonuria was completely obviated, and by its termination the patient was asymptomatic and able to eat. Recovery thenceforth was undisturbed and smooth.

With the description of these two cases we wish to stress the necessity of close observation for ketonuria throughout the preoperative, and the critical days of the postoperative periods, starting with the *very first* urine specimen obtained after the operation. Ketosis must be prevented, or if it emerges, it must be abolished at its very inception.

As regards the objections to 10 per cent glucose concentrations, first, we have not seen thrombosis any more frequently with this concentration than with weaker solutions.

For glucose solutions of any concentration to induce diuresis it is obvious that glycosuria must first obtain. Glycosuria is a consequence of excessive hyperglycemia which, in turn, is due to failure of the body to assimilate glucose at the rate at which it is infused.

In contemplating the merits of the view that 10 per cent glucose solution induces diuresis we must divide operative patients into two groups: the first comprising those with normal or near normal carbohydrate tolerance, and the second represented by known diabetics and such patients whose carbohydrate tolerance is lowered by the surgical disease or effects of the surgical

TABLE III.—*Showing that Diuresis Caused by Infusion of 1000 cc. of 10 Per Cent Glucose Is Insignificant Even When Moderate Hyperglycemia Occurs*

Patient Description	Time After Start of Infusion Hours	Blood Sugar (Venous Blood) Mg. Per Cent	Urine Volume cc.	Urine Sugar Gm.
B.M.*	0	98	...	...
Female, age 59, with minor fracture.	0.5	248	...	...
In apparent good health for age;	1	302	5	0.03
signs of generalized arteriosclerosis	2	337	50	0.4
apparent	Infusion ended			
	3	101	190	1.0
	4	65	70	0.4
J.K.**	0	88	...	0
Female, age 47, with toxic hepatitis	0.5	225	70	0.2
(proven by liver biopsy)	1	313	120	0.6
	2	379	150	1.2
	Infusion ended			
	3.5	...	260	2.6
	4.5	...	50	0.3
	5.5	...		

* 800 cc. of 10% glucose, administered at constant rate in the course of two hours (40 Gm. glucose per hour).
** 1000 cc. of 10% glucose, administered at constant rate in the course of two hours (50 Gm. glucose per hour).

procedure. In the first group, the production of diuresis by 10 per cent glucose solution is not possible so long as the rate of infusion does not exceed 50 to 60 Gm. of glucose per hour. This fact was first demonstrated by Woodyatt[7] who showed that, on an average, 0.85 Gm. of glucose per kilo bodyweight per hour can be administered intravenously to a healthy person without the production of glycosuria. This means that an average individual of 70 kilo bodyweight, whose carbohydrate tolerance is normal, does not develop abnormal hyperglycemia leading to glycosuria when glucose is administered intravenously at a rate of 600 cc. of 10 per cent solution per hour. And without glycosuria, no diuresis can result from such infusion. Besides,

in order to allow a safety margin, one should use a somewhat lower rate of infusion, as for example, 2.5 to 3 hours for 1000 cc. of solution. In Table II is presented an example in which under such conditions, no diuresis takes place.

But even when the infusion of 10 per cent glucose solution entails mild degrees of glycosuria, due to unforeseen impairment of the glucose tolerance, diuresis is still absent. This is clearly illustrated by the two examples recorded in Table III.

The glucose in these two instances was infused at rates of 40 and 50 Gm. per hour, respectively, and the urine was collected by catheterization during the infusion and for two and two-and-a-half hours after its termination. From the data in Table III it is evident that there was retention of fluid rather than diuresis, although the blood sugar in both cases rose well above 300 milligrams per cent and small amounts of glucose were excreted in the urine.

It is a significant fact, however, that the glucose tolerance of many patients is more or less impaired in the postoperative state. This condition may be of transitory nature, as proved in the course of our studies by entirely normal response to the conventional glucose tolerance test following surgical convalescence. In other instances we found distinct, though not severe, forms of diabetes which were not diagnosed before the operation probably because the disease was in an early stage in which glycosuria occurred only intermittently and was completely absent on a balanced hospital fare. Such latent diabetes is greatly exacerbated in the course of major surgical treatment and is subject to further degeneration if ketosis is allowed to develop.

These observations explain why we came upon considerable degrees of glycosuria when the rate of glucose infusion was as low as 30 Gm. per hour, although the patient was not diagnosed as diabetic. And it was such observations that led us to collect—as a regular routine—urines during and following parenteral glucose administration for the quantitative determination of glucose. While so doing we found instances in which 25 per cent to 50 per cent of the glucose was lost when infused at the rate of 50 to 60 Gm. per hour. Part of the glucose may be lost even when infused at the rate of 25 Gm. per hour in the form of 5 per cent solution. And this is exactly the condition in which the patient can least afford to lose any of the glucose supplied him. Furthermore, this is the condition in which glucose does cause diuresis. Thus, the actual problem is to infuse the glucose at a rate which is within the assimilatory power of the patient, regardless of whether it is given in 5 per cent or 10 per cent solution. The infusion may be too fast in relation to the tolerance of the patient even if a 5 per cent solution is used, and an appropriately low rate of infusion may be observed when using a 10 per cent solution.

The answer to the problem, then, is to infuse the glucose at a rate which is not in excess of the rate of assimilation. This does not mean, as might at first seem apparent, that the infusion must be extended over five or six hours when glycosuria indicates a low rate of assimilation. In most instances it is only necessary to keep the rate low during the first hour or even half-hour,

following which it can be safely increased to a rate of 50 Gm. per hour. This rule is based upon a well known physiologic law. Staub[8] and others have observed that if two doses of glucose are administered orally at one hour intervals, the second dose incites considerably less hyperglycemia than the first. This phenomenon is due to the fact that the hyperglycemia produced by the first dose stimulates the assimilatory mechanism,* with the result that the second dose of glucose is utilized at a much higher rate than the first.

TABLE IV.—*Showing the Increase in the Rate of Assimilation Under the Stimulus of Hyperglycemia.*

Time After Start of Infusion Hours	Venous Blood Sugar mg. %	Glucose in Extracellular Fluid* Gm.	Increment in Extracellular Fluid Gm.	Glucose Infused Gm.	Glucose Excreted Gm.	Glucose Assimilated Gm.	Assimilation rate Gm./hr.	Remarks
0	98	13.7	....	..	...	....	....	Case 1. Wt. 54.5 kilo
0.5	248	27.8	14.1	20	0	5.9	11.8	Infusion of 80 Gm. glu-
1	302	33.8	6.0	20	0.4	13.6	27.2	cose at the rate of 40
2	310	34.7	0.9	40	1.0	38.1	38.1	Gm./hr.
3	88	9.8	—24.9	0	0	24.9	24.9	
4	65	7.3	—2.5	0	0	2.5	2.5	
0	91	19.1	....	..	...	....	....	Case 2. Wt. 84 kilo
0.5	195	32.8	13.7	20	0	11.3	22.6	Infusion of 100 Gm. glu-
1	231	38.6	5.8	25	1.3	17.9	35.8	cose at the constant rate
2	204	42.8	4.2	50	0	45.8	45.8	of 50 Gm./hr.
3	74	12.4	—30.4	0	0	30.4	30.4	
4	78**	13.1	0.7	0	0	—0.7	—0.7	
0	89	16.0	....	..	...	....	....	Case 3. Wt. 72 kilo
0.5	165	23.8	7.8	14	0	6.2	12.4	Infusion of 100 Gm. glu-
1	150	21.6	—2.2	14	0	16.2	32.4	cose at the rate of 28 Gm.
2	180	25.9	4.3	72	0	67.7	67.7	in first hour, 72 Gm. in
3	60	8.6	—17.3	0	0	17.3	17.3	second hour. Perspired
4	81**	11.7	3.1	0	0	—3.1	—3.1	freely, felt "shaky" one hour after termination of infusion

* Free glucose is confined to the extracellular fluids, including the whole blood. There is no free glucose in the cell content, since the cells assimilate glucose only after its conversion into glucose phosphate. The distribution of glucose in the extracellular fluid is not quite even. The concentration is highest in blood plasma; in the corpuscles it is 15 to 20 per cent lower. We have estimated the average glucose concentration in the total extracellular fluid as roughly 20% lower than in the blood. We have taken the total amount of extracellular fluid as 25% of the total bodyweight. By use of these figures and knowing the venous blood sugar value, the amount of glucose in the extracellular fluid was calculated. From this, knowing the amount of glucose infused and the amount lost in the urine in a fixed period of time, the amount of glucose assimilated was determined.

** The increase in blood sugar (glucose in extracellular fluids) is a compensatory response to hypoglycemia. This glucose is produced by dissimilation (phosphorylation with subsequent dephosphatization) of hepatic glycogen.

This law is equally valid, as we have observed in many experiments, when glucose is administered parenterally. In Table IV, three cases are presented which demonstrate this fact.

CASE REPORTS

Case 1 is a 63-year-old woman with an essentially negative past history, hospitalized for a relatively minor fracture; there were moderate evidences of generalized

arteriosclerosis. An intravenous infusion of 10 per cent glucose solution was given the patient at the constant rate of 40 Gm. per hour. As may be seen, the assimilatory rate (Gm./hr.) was 11.8 in the first half hour, 27.2 in the second half hour, and 38.1 in the second hour of infusion. In Case 2, a robust man of 23 years with acute cellulitis of the face, a similar infusion was administered at the constant rate of 50 Gm. per hour. The rate of assimilation (Gm./hr.) was 22.6 during the first half hour, 35.8 during the next half hour, and to 45.8 during the second hour. Both of these cases show an impaired glucose tolerance, as indicated by the blood sugar values determined in the venous blood.

Case 3 (Table IV) is a robust, healthy man of 21 years, who showed no evidence of impaired carbohydrate metabolism. His only pathology was a very minor fracture, and he seemed suitable for the subject of an experiment to demonstrate more emphatically the acceleration of the assimilatory mechanism under the stimulus of hyperglycemia. He was given an intravenous infusion of 10 per cent glucose solution at an initial rate of 28 Gm. per hour, the rate being increased to 72 Gm. per hour at the beginning of the second hour. As may be seen in the table, assimilation of the glucose started rather sluggishly, so that only 6.2 Gm. of glucose was assimilated in the first half hour (12.4 Gm./hr.) and the venous blood sugar reached a level of 165 mg. per cent. Under the stimulus of this hyperglycemia the rate of assimilation rose to 32.4 Gm./hr. in the second half hour, while in the second hour, when the glucose was being infused at a rate almost treble that of the first hour, the assimilatory rate was doubled to 67.7 Gm./hr., and the venous blood sugar rose only to 180 mg. per cent.

These observations prompt a quantitative procedure for the administration of glucose. The intravenous infusion of 10 per cent solution is begun at a slow rate (20 to 25 Gm.) and accelerated to 50 to 60 Gm. per hour after the first half hour at which rate it is continued until completed. All urine specimens obtained during, and for several hours following, infusions are quantitatively analyzed for sugar content by a simple method which requires no more skill and time than that necessary for the adequate execution of a qualitative test.[9] In those patients who, at any time, show more than minimal glycosuria, and in those who are suspected of markedly depressed glucose tolerance due to any of the causes previously discussed, serial, quantitative urine sugar determinations are made at frequent intervals during the course of infusion, and the rate of infusion not accelerated until there is no, or negligible, glycosuria.

Moderate glycosuria, which represents less than 10 per cent of the infused glucose, does not invite serious misgivings as to the correctness of the procedure. It still represents fair economy and, as shown in Table III, does not entail diuresis. It is always desirable, however, to determine quantitatively the extent of glycosuria during and for two hours after the infusion in order to ascertain that there be no undue waste of glucose.

If the loss through glycosuria amounts to 10 per cent or more of the glucose infused, despite the precautions just described, the use of insulin is

* The term "assimilation" is used here to denote the process of incorporation of blood sugar into the cell content. This process is contingent upon the phosphorylation of glucose, since cells are impermeable to free glucose. The phosphate esters of glucose are then either oxidized in the cells or polymerized to glycogen for storage. The latter processes, however, are not part of the process of assimilation; the former is a storing process, the latter catabolism.

indicated. The same holds, of course, for diabetic patients. Insulin in these cases is of great service, but can do as much harm as good if it is not governed by due consideration to certain physiologic principles. We intend to discuss this aspect of the problem in a separate note.

SUMMARY

1. The purpose of postoperative parenteral glucose therapy is two-fold: prevention of starvation ketosis, and exertion of its protein-sparing effect.

2. This dual purpose can be achieved with amounts of glucose no less than 200 to 350 Gm., well spaced over the 24 hours.

3. Such amounts of glucose, if they are administered in solutions of less than 10 per cent concentration, make the total amount of fluid excessive, and often prohibitive. The concentration of the glucose solution, therefore, must not be less than 10 per cent.

4. The view that 10 per cent solutions necessarily cause diuresis is erroneous. If the solution is infused at proper rates, in keeping with the rate of assimilation, excessive hyperglycemias and consequent glycosurias are precluded and hence diuresis does not occur.

5. A rational procedure of intravenous glucose infusion entails an initial low rate of infusion of 10 per cent solutions (in combination with Ringer's solution or physiologic saline if desired), with subsequent acceleration of the rate. Serial quantitative determinations of urine sugar show whether or not the rate of infusion exceeds the rate of assimilation and thus serve in guiding the regulation of the rate of infusion.

BIBLIOGRAPHY

[1] Wells, H. G.: Arch. Int. Med., **1**: 589, 1908: Chemical Pathology, 3rd Edition. Philadelphia, Saunders, 1918.
[2] Opie, E. L., and L. B. Alford: J. A. M. A., **62**: 895, 1914; J. Exper. Med., **21**: 1, 1915.
[3] Graham, E. A.: J. Exper. Med., **21**: 185, 1915.
[4] Davis, N. C., and G. H. Whipple: Arch. Int. Med., **23**: 612, 1919.
[5] Miller, L., and G. H. Whipple: Am. J. Med. Sci., **199**: 204, 1940.
[6] Probstein, J. G., and N. W. Drey: J . Mo. State Med. Assoc., **37**: 475–477, 1940.
[7] Woodyatt, R. T., W. D. Sansum, and R. M. Wilder: J. A. M. A., **65**: 2067, 1915.
[8] Staub, H.: Ztschr. f. Klin. Med., **93**: 89, 1922.
[9] Somogyi, M.: J. Lab. & Clin. Med., **26**: 1220, 1941.

DISASTERS FOLLOWING THE OPERATION OF LIGATION AND RETROGRADE INJECTION OF VARICOSE VEINS

JOSEPHUS C. LUKE, M.D., F.R.C.S. (ENG.) & (C.)
AND
G. GAVIN MILLER, M.D., F.R.C.S. (C.)
MONTREAL, CANADA

FROM THE DEPARTMENT OF SURGERY OF THE ROYAL VICTORIA HOSPITAL AND MC GILL UNIVERSITY, MONTREAL.

THE OPERATION of high ligation and retrograde injection of the saphenous vein has become the treatment of choice in those cases of varicose veins showing evidence of retrograde flow. This appears to be a rational method for, in the presence of incompetent valves, the weight of the column of blood from the femoral ring to the ankle interferes with venous return in the lower leg, and leads to pooling of blood in the large varicose veins.

It appears to be the belief of the occasional operator that this is a simple, easily carried out procedure, free from hazard. Consequently, the operation is widely practised by inexperienced surgeons and hospital internes.

As might be expected unfortunate results are occurring with increasing frequency. The purpose of this paper is to report 21 such cases, and, from a study of the results, to offer suggestions which might decrease the incidence of complications. It is quite obvious that minor surgical complications, such as haematomata and infections, will occur in a percentage of these cases depending on the care and skill with which the operation is carried out. However, we wish only to discuss the serious disasters such as occlusion of the artery by operative trauma, deep femoral phlebitis, and, perhaps, those more frequently unfavorable results due to damage caused by sclerosing solutions. It should also be mentioned that all these adverse results happened not only to occasional, or junior, operators, but also to senior surgeons of great experience and high standing.

The operation of ligation of the great saphenous at the saphenofemoral junction may be difficult in an obese individual with a thinned out varix near the terminal portion, especially if, due to previous phlebitis and periphlebitis, firm adhesions are present. The varix is easily torn, or one of the large branches may escape the ligature. It should be appreciated that very little pressure is required to stop this flow of blood but, at the moment, the unexpected severity of the bleeding upsets the calm of the surgeon and uncontrolled clamping results in injury to the deep femoral vein, the artery or the nerve.

Case 1.—While freeing a large varicose saphenous vein a large tributary was torn near the terminal portion. Kelly clamps were hurriedly applied which, on later investigation, proved to have included the femoral vein necessitating its ligation. When last seen some months later marked oedema of the leg was present. This patient, a soldier, had to be re-categorised and returned to base duties. On discharge he was found to be pensionable because of this permanent disability.

Case 2.—This patient was operated upon, under local anaesthesia, by a skilled surgeon in charge of a varicose vein clinic, in the Outdoor Department of a large teaching hospital. She was obese, a known hypertensive, and an electrocardiograph had shown evidence of myocardial degeneration. Her veins were large, troublesome and showed valvular incompetence. The past history revealed an episode of phlebitis. At operation, a large varix was present at the saphenous junction, adherent to the adjacent tissues. While dissecting this free, the saphenous was torn at its femoral junction. Severe hemorrhage occurred which could not be arrested by careful clamping. Finally, bleeding was controlled by pressure while a blood transfusion was given and a general anesthetic commenced. An enlarged incision was made to allow dissection of the femoral vein, which was ligated. However, the patient went into shock, and, in spite of blood transfusion, died half an hour later. Autopsy showed no lesion to account for her death, so it must be presumed that this severe hemorrhage overtaxed a damaged cardiovascular system, with fatal result.

After such an accident it is easy to point out how the hemorrhage could have been more safely handled, for example, by the use of packing and a pressure bandage, but these emergencies occur quickly and become frightening to the surgeon, who usually makes every effort to arrest the bleeding by clamping the bleeding point.

In this presentation no example of ligation of the femoral vein with retrograde injection of sclerosing fluid is included, but when it is realized how superficial the femoral vein is in the inguinal region, it is easy to imagine that the saphenous may be considered as a tributary rather than as the main vein, and the femoral ligated and injected in consequence. Such an accident would certainly result in a marked oedema, and later in a so-called postphlebitic leg.

Case 3.—This patient had had a previous ligation and injection of the saphenous vein with recurrence of varicosities. A surgeon, with considerable interest and experience in the treatment of varicose veins, elected to explore the saphenofemoral junction The adhesions from the former operation made the procedure difficult but, presumably, the vein was found, ligated and distally injected with sodium morrhuate. Immediate pallor and paresis of the leg occurred. When the wound was reopened, it was found that the femoral artery was ligated, divided and distally injected. In spite of palliative treatment, a mid-thigh amputation was required.

Case 4.—A young woman underwent ligation and injection of the saphenous, under local anesthesia. When the vein was injected, severe pain occurred from the groin to the toes, the leg blanched, and no arterial pulsation was obtainable. This patient was seen by one of us in consultation. The leg was found to be cold, blanched, exceedingly sensitive and pulseless. Subsequent gangrene necessitated a mid-third thigh amputation. Histological examination showed thrombosis of the major arteries. The surgeon stated that the vessel he ligated did not pulsate, but it is well recognized that a state of spasm often occurs in a vessel on exposure and handling which minimizes pulsation. In this case, the original wound was not explored. The young, but well-trained, surgeon is now defending a lawsuit for very large damages and has suffered a serious setback in his career.

Case 5.—This patient underwent bilateral saphenous ligation and injection. Eight cc. of sodium morrhuate solution were injected into both veins. Considerable pain was felt in the left thigh after the operation. The veins were well thrombosed and the leg arteries were noted to be pulsating normally on discharge three days later. A month later, she was readmitted with a sloughing, gangrenous, foul-smelling ulceration, five

inches in diameter, in the left upper posterior aspect of the thigh. Bacterial study revealed anaerobic streptococci as the predominant organism. On excision, it was found that the gangrenous slough involved the surface of the femur, included the sciatic nerve, and extended into the perineum. Amputation was considered but refused. The femur has now regenerated and the wound has healed but sciatic paralysis remains. The Wassermann is negative, the blood sugar curve is normal, and arterial circulation has always been adequate. This operation was carried out by a senior surgeon of great experience and there seems little possibility that any technical error occurred. The area of tissue necrosis corresponds to the distribution of the profunda femoris artery, and it has been suggested that the complication was due to spasm of this artery. There are many objections to this explanation.

Case 6.—This patient underwent a high and low ligation of the right saphenous, under general anesthesia. Sodium morrhuate was injected at the time of operation. The patient was confined to bed for three days because of fever. Six days after the first operation the left saphenous was ligated and injected. Because of fever the patient was again confined to bed. Six days later, still febrile, she complained of a tight feeling in the chest, general malaise and a dry cough. A chest roentgenogram revealed a shadow suggestive of pneumonia at the right base. A second acute episode occurred eight days later. One month following her second operation she died suddenly. Autopsy showed thrombosis of all the great veins of the left leg extending up to the inferior vena cava, with healing infarcts in the lower lobe of the right lung, and massive recent emboli filling the pulmonary artery.

Case 7.—This patient, a young woman, died suddenly at home, a week after ligation and injection of the left saphenous vein, presumably from a pulmonary embolus.

Case 8.—This patient had bilateral saphenous ligation. Four days later, daily injections of calf varices with 2 cc. of sodium morrhuate were begun. He was discharged two weeks later but developed a pulmonary embolus shortly after leaving the hospital. Two further emboli occurred associated with bilateral pleural effusion. The patient had bilateral thrombophlebitis. He was discharged from the Army, and when recently examined showed bilaterally swollen, aching legs, with medial supramalleolar ulcers. In this case, the saphenous veins having been ligated, the phlebitis presumably occurred in the deep femoral vein.

In all, information has been received of four fatalities and ten further cases of deep thrombophlebitis following ligation and injection of saphenous veins.

The cases of deep thrombophlebitis all have permanent disability, some with persistent oedema, eczema and ulceration of the leg. The cases which occurred in the Services will all be pensionable.

It may be true that operative accidents, as described above, are comparatively rare. Two other cases of femoral artery ligation are known to the authors, but not reported here because of insufficient data. What has been reported, however, should be sufficient to show the hazards of saphenous ligation and retrograde injection, and to emphasize the difficulties the procedure may present to the surgeon. In order to save beds, many hundreds of these cases have been done in the outpatient clinic of the Royal Victoria Hospital in the Varicose Vein Clinic, under local anesthesia. This procedure, except for the death reported previously, has proved satisfactory,

especially as immediate ambulation appears to be of great value. However, certain rules have had to be laid down. Adequate operating room equipment and nursing staff must be available, together with an assistant of some experience. The operation can only be carried out when the senior surgeon in charge of the clinic is in attendance in the outpatient clinic. In the event of any unusual complication the advisability of admitting the patient, or of immediately calling one of the senior consultants, must be considered. The surgeon must be familiar with the anatomy of the saphenous vein (and especially its tributaries), the fossa ovalis and its relation to the deep fascia, and also the relationship of the saphenous to the femoral vein and artery. A useful anatomic consideration is the fact that the superficial external pudendal artery, though occasionally variable, runs on the lower superficial edge of the fossa ovalis posterior to the incurving saphenous vein. It is, therefore, a good guide to the sapheno-femoral junction. After incising the skin, one should feel carefully for the pulsation of the femoral artery, so that adequate orientation may be achieved. Dissection must be meticulous. In this way will the operative hazards be reduced to a minimum.

If the main vein is torn, blind clamping is dangerous. The bleeding is venous, of low pressure, and can easily be controlled by the finger or by packing. Lowering the head of the table still further lessens the pressure. For this reason it is a wise precaution to fix shoulder rests on the table before operating. Suction apparatus should always be immediately available in case of bleeding. Careful removal of the packing after several minutes with the use of suction to keep the wound free of blood, usually shows the point of bleeding which can be delicately clamped. Ligation of the femoral vein, which has usually been torn, is then indicated.

It is still open to question whether primary femoral arterial thrombosis can occur as a result of saphenous vein ligation and injection. It appears possible, however, that transient arterial spasm may result. The authors have observed five such cases out of 756 operations carried out under local anesthesia. In these cases greater pain than usual occurred in the leg immediately following operation. The pain was of a burning cramp-like character. In three cases the pain radiated to the lumbar region and necessitated temporary discontinuance of the operation. The leg blanched and no pulsation could be felt in the involved foot. The pain disappeared in thirty minutes. The only reasonable explanation appears to be that operative trauma, adjacent to the femoral artery, produced spasm sufficient to arrest the blood flow for half an hour.

The majority of accidents are due to deep venous thrombosis and embolism. When large quantities of sclerosing solutions are injected into an incompetent vein some of the fluid is apt to enter the deep system through communicating branches. Every surgeon has noted the complaint of the patient of a bitter taste in the mouth, or of an abdominal cramp following the injection of 2 cc. of quinine urethane into an incompetent superficial varicosity. Presumably, this fluid has passed into the deep venous circulation, and been

absorbed. Perhaps this phenomenon is illustrated in that considerable group of patients who show persistent oedema following saphenous ligation and injection. In the rare cases who die of embolism, other unsuspected factors may be present; for example, an increased clotting tendency, the use of a larger amount of sclerosing solution than is usually advised, or prolonged post-operative inactivity.

The authors have collected 16 accidents due to thrombosis, but as these cases have been seen not only in our own hospital but also in the Services and in consultation, no percentages can be arrived at. Perhaps this is just as well for the percentage would be low enough to give the surgeon a feeling of safety, but as the widow said, "The percentage might as well be 100 per cent as far as my husband and I are concerned." Of the 16 cases referred to, with deep venous thrombosis following ligation and injection, four produced fatal pulmonary embolism, and two developed minor emboli.

Against the facts of this report must be weighed the knowledge that the ligation and retrograde injection treatment of varicose veins appears to be the best yet evolved. It is not our wish to discredit it. We have observed that the thrombotic accidents all occurred in patients who did not walk for 12 hours or more following operation. In several cases general or spinal anesthetics had been used which delayed walking for several hours. It is suggested, therefore, that only a local anesthetic be used, and that patients walk immediately after operation. Further, it was noted that in all these cases more than 6 cc. of sclerosing fluid were used. It is suggested that no more than 5 cc. of any sclerosing solution be injected at the time of operation. Experience may finally show that ligation alone should be carried out, with later injections once a week until the varicosities have been obliterated.

In a series of 756 ligations and injections, mostly on outpatients, under local anesthesia, who walked immediately after operation and for 15 minutes every hour following operation with active dorsi—and plantar flexion of the foot as soon as the sclerosing solution had been injected, we have had no case of embolism. Early ambulation is designed to ensure a rapid flow of blood in the deep venous system. In none of these cases has more than 6 cc. of 5 per cent sodium morrhuate been injected, and, by drawing back blood into the syringe, this amount has been diluted three to one with blood before injection. One death occurred as reported above (Case 2) due to actual operation. One case had a deep phlebitis, treated by subsequent ligation of the superficial femoral vein.

If an embolus should occur, we advocate removal of the clot in the vein with ligation of the superficial femoral vein. This appears safer than the use of dicoumarol, but, if ligation is not feasible, dicoumarol in adequate doses should be given, controlled by frequent estimations of the prothrombin time. Again, it should be emphasized that this report is not intended to discourage the use of the operation of ligation and injection of the saphenous vein for varicose veins with incompetency, but to warn of the dangers and offer a few suggestions as to how these dangers may be overcome.

SUMMARY

1. This paper is presented to emphasize the increasing number of untoward results following the operation of ligation and retrograde injection of varicose veins. Twenty-one such cases are referred to.

2. These disasters belong to two main groups, those resulting from operative difficulties and mistakes, and those resulting from deep venous thrombosis subsequent to operation. The latter group is the larger.

3. Study of the thrombotic group shows that two factors probably play a considerable role in the formation of this thrombosis. These are lack of muscular activity of the legs following operation, resulting in a slowing of the deep venous return, and the use of too great a quantity of sclerosing fluid.

4. Suggestions are made with a view to preventing the disasters of both the operative and the thrombotic groups.

SACROCOCCYGEAL CHORDOMA

FERNANDO GENTIL, M.D.
RIO DE JANEIRO
AND
BRADLEY L. COLEY, M.D.
NEW YORK, N. Y.

FROM THE BONE TUMOR SERVICE, MEMORIAL HOSPITAL, NEW YORK

CHORDOMA, though a rare tumor, is encountered often enough to require consideration, particularly in a tumor clinic where there are other types of neoplasms from which it must be differentiated. For those interested in oncology a knowledge of the clinical course, diagnosis and prognosis of chordoma as well as its response to various therapeutic methods is important. This study is based on an analysis of seven cases observed and treated at the Memorial Hospital from 1930 to 1943, inclusive; in addition, 128 previously reported cases have been reviewed.

Definition. Chordoma is a specific tumor arising from the remnants of the primitive notochord. It is characterized by slow growth which is inexorably progressive, a tendency to invade and destroy bone by direct extension, local recurrence after surgical excision and slight or negligible regression following irradiation. Occasionally chordoma is capable of regional and distant lymph node and visceral metastases.

Historical Summary. Attention was first called to chordoma in 1856 by Virchow[93] who described small tumor-like masses at the spheno-occipital synchondrosis. It was Virchow's belief that this tumor was essentially cartilaginous in nature and he called it *ecchondrosis* (from cartilage) *physaliphora* (vacuole containing cell). Two years later Muller[64] suggested that there was a relationship between this growth and the chorda dorsalis and accordingly introduced the term chordoma (chordoid tumor). It was not until 1894 that the true nature of this neoplasm was definitely established by Ribbert[78] who reported five cases of his own. This author carried out extensive morphologic investigations and the evidence which he accumulated supported Muller's notochordal origin of chordoma. In addition, Ribbert is credited with demonstrating the derivation of chordoma from notochordal remnants by puncturing the nucleus pulposus in rabbits and producing a tumor which was supposed to be grossly and microscopically identical with chordoma. The first case of sacrococcygeal chordoma was described in 1900 by Henning[42] who encountered this tumor while performing a postmortem examination on a seven-months-old stillbirth.

Although several comprehensive reviews of the literature have been published by Stewart, Mabrey, and others, most of the reports have been based on personal experience with only a single or a few cases. Aside from the

present report, only three clinical pathologic studies have been made based on a relatively large series of chordoma in the sacrococcygeal region, namely, that of Seneque and Grinda (8 cases); Fletcher, *et al* (10 cases); and Mabrey (8 cases). In a complete review of the literature on sacrococcygeal chordoma we found only 128 reported cases, or a total of 135 including the seven reported by us.

Incidence. It is difficult to determine with accuracy the incidence of chordoma since it occurs so rarely. During the period from 1930 to 1943 inclusive, 31,099 cases of cancer were seen at Memorial Hospital (first admission) and among these were only seven cases of sacrococcygeal chordoma and two of the spheno-occipital type.

Age. Although chordomas may occur at any age, they are usually encountered in about the 5th decade of life. Of the 128 cases reported in the literature (Table I), the youngest patient was three months old, the oldest 78 years of age, and the median age was 46 years. In the series herein reported (Table II), the youngest patient was 50 years and the oldest 70 years of age; the median age was 60 years. It is likely, however, that chordoma arises at a much earlier age than these figures reveal for an analysis of our own and previously reported cases shows that there is an average interval of about three years between the onset of symptoms and hospital admission. Even the age of the onset of symptoms is not a reliable index of the date of origin of the tumor for chordoma is admittedly slow growing and in all likelihood arises many years before the appearance of clinical manifestations.

Sex. Sacrococcygeal chordoma is encountered more often in male than in female patients. In the present series of seven cases there were five males and two females and of the 128 patients reported in the literature 68 per cent were males. The significance of the predominance of males is difficult to evaluate, but there is no evidence to support the thesis that this neoplasm might be considered one of the sex-linked tumors.

ETIOLOGY

Trauma. The question of trauma as an etiologic factor in the production of certain anatomic forms of cancer is complicated and highly controversial. A recent excellent review of this subject by Stewart[86] indicates that, at least in the human, single or multiple injuries, especially in the case of a bone tumor, cannot be seriously regarded as a significant factor in the etiology of neoplasms. Although Ribbert was supposed to have produced experimental chordoma in the rabbit by puncturing the nucleus pulposus, his work is inconclusive and has never been confirmed. A history of trauma was obtained in two of our seven cases, and of the previously reported cases of sacrococcygeal chordoma in the literature, definite trauma to the lower end of the spine was recorded in 32 per cent. It is our conclusion that the relationship of injury to the onset of this neoplasm remains unproven.

Development Anatomy. The anatomic origin of the notochord has never been definitely established. We do know, however, that at an early period of

TABLE I.—*Sacro-Coccygeal Chordomas.* Cases Collected from the Literature

No.	Author	Year	Sex	Age	History of Trauma	Histol. Diagn. Section-Aspiration	Duration of Symptoms	Treatment: Radiation	Treatment: Surgery	Recurrences	Metastasis	Survival Period	End Result
1.	Henning	1900	M	7 mos.	No	Section	.....	...	...	...	No	.....	.....
2.	Mazzia	1910	M	54	No	Section	?	No	Yes	Yes	No	1 year 7 months	?
3.	Feldman	1910	M	46	No	Section	1 year	No	Yes	Yes	No	2 years	?
4.	Wagner	1910	M	68	No	Section	6 years	No	Yes	Yes	Lymph nodes	1 year	?
5.	Curtis	1911	M	58	No	Section	6 months	No	Yes	Yes	No	2 years	Dead of disease
6.	Wood	1913	F	35	No	Section	?	No	Yes	Yes	No	4 years	?
7.	Dibernardi	1913	M	56	No	Section	2 months	No	Yes	?	No	?	?
8.	Albert	1915	M	26	Yes	Section	3 months	No	Yes	Yes	No	14 months	Dead of disease
9.	Tuffier & Vignes	1918	F	55	No	Section	3 years	No	Yes	Yes	No	3 years	?
10.	Lund	1919	F	60	No	Section	3 years	No	Yes	Yes	No	1 year 2 months	Dead of disease
11.	Pototschnig	1919	M	40	No	Section	10 years	No	Yes	...	Lymph nodes	.....	Died postoperatively
12.	Peters	1919	M	68	No	Section	5 years	No	Yes	Yes	Lymph Nodes	?	Dead of disease
13.	Wiethold	1920	M	61	No	Section	?	No	Yes	?	?	?	?
14.	Lewis	1921	M	54	Yes	Section	?	?	?	?	?	?	?
15.	Lewis	1921	M	30	No	Section	?	?	?	?	Lymph nodes and liver	?	Dead of disease
16.	Lewis	1921	M	58	No	Section	2 years	No	Yes	?	Lymph nodes	?	?
17.	Lewis	1921	F	22	No	Section	2 years	No	Yes	Yes	Lymph nodes Peritoneum	1 year	Dead of disease
18.	Stewart	1922	M	65	No	Section	8 years	No	Yes	Yes	Skin and subcutaneous tissue	11 years	Dead of disease
19.	Linck	1922	M	61	No	Section	2 years	Yes	Yes	Yes	No	?	?
20.	Micotti	1922	M	53	Yes	Asp. biopsy	?	No	Yes	Yes	No	6 months	Dead of disease
21.	Beard, et al	1922	M	22	Yes	Section	1 year	Yes	Yes	Yes	No	1 year 6 months	Dead of disease
22	Pool	1922	F	65	No	Section	2 years	No	Yes	Yes	No	?	?
23.	Andler	1923	M	45	Yes	Section	3 months	No	Yes	Yes	No	1 year	Dead of disease
24.	Andler	1923	M	62	No	Section	5 years	No	Yes	...	.....	.....	?
25.	Hirsch & Ingals	1923	M	54	No	Section	1 year 8 months	No	Yes	No	No	3 years	Dead of disease
26.	Raul	1924	M	49	No	Section	6 months	No	Yes	...	.....	.....	?

TABLE I.—(*Cont'd*) *Sacro-coccygeal Chordomas.* Cases Collected from the Literature

No.	Author	Year	Sex	Age	History of Trauma	Histol. Diagn. Section-Aspiration	Duration of Symptoms	Treatment: Radiation	Treatment: Surgery	Recurrences	Metastasis	Survival Period	End Result
27.	Arguad	1925	F	14 mos.	No	Section	?	No	No	...	Liver	?	?
28.	Walz	1925	M	62	No	Section	6 months	No	Yes	...	.....	.....	Dead of disease
29.	Hayman	1925	F	60	No	Section	1 year	No	Yes	No	No	1 year	?
30.	Young	1925	M	49	No	Section	3 years 6 months	No	Yes	Yes	No	2 years 6 months	?
31.	Argaud	1925	F	48	Yes	Section	7 months	No	Yes	?	?	?	?
32.	Richardson & Taylor	1926	M	43	No	Section	6 months	No	Yes	...	?	?	Dead of disease
33.	Kersner	1926	M	45	No	Section	2 years	No	Yes	?	?	?	?
34.	Sommer	1926	M	56	No	Section	?	No	Yes	?	?	?	?
35.	Andler	1926	F	57	No	Section	2 years	No	Yes	?	?	?	Dead of disease
36.	Andler	1926	M	50	Yes	Section	3 months	Yes	Yes	Yes	No	1 year	Dead of disease
37.	Andler	1926	M	49	No	Section	2 years	No	Yes	No	No	1 year	?
38.	Stewart & Morin	1926	M	58	No	Section	6 months	No	Yes	...	No	33 years	?
39.	Alexander & Struthers	1926	M	64	No	Section	1 year 6 months	No	Yes	...	No	1 year 6 months	?
40.	Cameron	1926	F	59	No	Section	1 year 6 months	No	No	...	No	?	?
41.	Kwartin & Stewart	1927	M	56	No	Section	1 year	Yes	Yes	No	No	2 years 4 months	?
42.	Letulle & Dujarier	1927	M	32	No	Section	?	No	Yes	Yes	No	7 months	?
43.	Reynes & Rouslacroix	1927	M	68	Yes	Section	20 years	Yes	Yes	Yes	No	?	Dead of disease
44.	Fumegalli	1927	F	68	No	Section	1 year	No	Yes	?	?	?	?
45.	Bustos	1928	M	67	Yes	Section	5 months	No	No	...	No	?	Dead of disease
46.	Podlaha & Pavlica	1928	F	44	No	Section	?	Yes	Yes	Yes	No	2 years	Dead of disease
47.	Conway	1929	F	20	No	Section	3 years	No	No	...	Lymph nodes Peritoneum Muscle Bladder Omentum	?	Dead of disease
48.	Tempsky	1929	M	52	Yes	Section	2 years	No	Yes	No	No	10 months	?
49.	Hutton	1929	M	49	Yes	Section	3 years 6 months	Yes	Yes	Yes	No	3 years	?
50.	Machulko & Rochlin	1930	M	57	No	Section	6 years	Yes	Yes	Yes	No	16 years	Dead of disease

TABLE I.—(*Cont'd*) *Sacro-coccygeal Chordomas.* Cases Collected from the Literature

No.	Author	Year	Sex	Age	History of Trauma	Histol. Diagn. Section-Aspiration	Duration of Symptoms	Treatment		Recurrences	Metastasis	Survival Period	End Result
								Radiation	Surgery				
51.	Gerber	1930	M	55	No	Section	2 years	No	Yes	...	No	5 days	Dead of disease
52.	Seneque & Grinda	1930	F	60	Yes	Section	1 year	No	Yes	Yes	No	9 years	?
53.	Seneque & Grinda	1930	M	39	Yes	Section	2 years	Yes	No	Yes	No	1 year	?
54.	Seneque & Grinda	1930	M	56	No	Section	6 months	Yes	No	Yes	No	?	?
55.	Seneque & Grinda	1930	M	78	Yes	Section	6 months	Yes	No	Yes	No	1 year	Dead of disease
56.	Seneque & Grinda	1930	M	32	Yes	Section	?	Yes	No	Yes	No	2 years	Dead of disease
57.	Seneque & Grinda	1930	M	63	No	Section	?	Yes	Yes	Yes	No	7 years	Dead of disease
58.	Seneque & Grinda	1930	F	68	No	Section	2 years	Yes	No	Yes	No	8 months	Dead of disease
59.	Seneque & Grinda	1930	F	53	No	Section	?	Yes	Yes	Yes	No	4 years	?
60.	Willis	1930	F	36	No	Section	2 years	No	No	...	Spleen, liver, lung, heart, kidney, skin thyroid	.....	Dead of disease
61.	Dickson	1931	M	41	No	Section	10 months	Yes	Yes	Yes	No	8 months	?
62.	Wildbolz	1931	M	52	Yes	Section	2 years	Yes	Yes	Yes	No	4 years	Dead of disease
63.	Sandahl	1932	M	23	No	Section	5 months	Yes	Yes	Yes	No	5 months	Dead of disease
64.	Wahling	1932	M	56	Yes	Section	3 years	No	Yes	...	No	.....	Dead of disease
65.	Stanton	1932	M	62	Yes	Section	2 years	Yes	Yes	Yes	No	?	?
66.	Estella	1932	F	3	No	Section	3 years	No	Yes	Yes	No	?	?
67.	Estella	1932	F	8	No	Section	1 month	No	No	No	No	.....	Dead of disease
68.	Cato	1932	M	35	Yes	Section	?	Yes	Yes	Yes	Liver	?	Dead of disease
69.	Pai	1932	M	62	Yes	Section	2 years	No	Yes	Yes	No	1 year	Dead of disease
70.	Chesky	1932	M	53	Yes	Section	1 year 6 months	No	Yes	No	No	1 year	?
71.	Grandclaude, et al	1933	F	36	No	Section	2 years	Yes	No	Yes	No	?	Dead of disease
72.	Montgomery & Wolman	1933	F	3	Yes	Section	2 months	No	No	...	Lungs	.....	Dead of disease
73.	Montgomery & Wolman	1933	F	3 mos.	No	Section	1 month	No	Yes	Yes	No	23 days	Dead of disease
74.	Montgomery & Wolman	1933	F	2	No	Section	1 month	Yes	Yes	...	No	?	?
75.	Donati & Manginelli	1933	M	53	No	Section	?	No	Yes	?	?	?	?
76.	Penkert	1933	F	28	No	Section	2 years	No	Yes	?	No	7 months	?
77.	Penkert	1933	F	34	No	Section	5 years	No	Yes	?	No	4 months	?
78.	Cordovil	1934	M	55	Yes	Section	?	Yes	Yes	Yes	No	10 months	?

TABLE I.—(*Cont'd*) *Sacro-coccygeal Chordomas.* Cases Collected from the Literature

No.	Author	Year	Sex	Age	History of Trauma	Histol. Diagn. Section-Aspir tion	Duration of Symptoms	Treatment		Recur-rences	Metastasis	Survival Period	End Result
								Radia-tion	Surgery				
79.	Fletcher, et al	1935	M	33	No	Section	2 years	Yes	Yes	Yes	No	9 years	Dead of disease
80.	Fletcher, et al	1935	M	68	No	Section	?	No	Yes	Yes	No	1 year	Dead of disease
81.	Fletcher, et al	1935	F	43	No	Section	8 months	Yes	No	Yes	No	3 months	Dead of disease
82.	Fletcher, et al	1935	F	25	Yes	Section	2 years	No	Yes	Yes	No	4 years	Dead of disease
83.	Fletcher, et al	1935	M	47	No	Section	2 years 6 months	Yes	Yes	Yes	No	3 years	Dead of disease
84.	Fletcher, et al	1935	F	65	Yes	Section	?	No	Yes	Yes	No	3 years	Dead of disease
85.	Fletcher, et al	1935	M	47	No	Section	1 year	Yes	Yes	Yes	No	4 years	Dead of disease
86.	Fletcher, et al	1935	M	58	No	Section	6 months	Yes	Yes	No	No	9 years	?
87.	Fletcher, et al	1935	M	61	No	Section	8 months	Yes	No	?	No	?	?
88.	Fletcher, et al	1935	M	25	No	Section	1 year	No	Yes	Yes	No	?	?
89.	Pupo, et al	1935	M	26	No	Section	?	No	Yes	Yes	No	?	?
90.	Coulthard & Harris	1935	F	49	No	Section	2 years	Yes	No	Yes	No	1 year	Dead of disease
91.	Coulthard & Harris	1935	M	53	No	Section	1 year	Yes	No	Yes	No	?	?
92.	Riche, et al	1935	M	33	Yes	Section	5 years	Yes	Yes	Yes	No	3 years	Dead of disease
93.	Mabrey	1935	F	52	No	Section	7 years	No	Yes	?	No	?	?
94.	Mabrey	1935	F	53	Yes	Section	5 years	No	Yes	?	No	?	?
95.	Mabrey	1935	M	38	Yes	Section	2 months	Yes	Yes	Yes	No	1 year 2 months	?
96.	Mabrey	1935	F	33	No	Section	2 months	Yes	Yes	Yes	No	4 months	Dead of disease
97.	Mabrey	1935	M	47	Yes	Section	6 months	No	Yes	Yes	No	2 years 6 months	Dead of disease
98.	Mabrey	1935	F	43	Yes	Section	4 years	Yes	No	?	No	?	Dead of disease
99.	Mabrey	1935	M	35	Yes	Section	1 year	Yes	Yes	No	No	3 years	?
100.	Mabrey	1935	M	42	Yes	Section	2 years	Yes	Yes	Yes	No	1 year 6 months	?
101.	Harmos & Palmer	1936	M	71	Yes	Section	4 years	Yes	Yes	Yes	No	?	?
102.	Gabriel	1936	M	49	No	Section	2 years	No	Yes	Yes	No	?	?
103.	Monserrat & Olascoaga	1937	M	1½	Yes	Section	1 year 3 months	Yes	No	Yes	No	?	Dead of disease
104.	Nash & Laskey	1937	M	54	No	Section	1 year	No	Yes	?	No	?	?
105.	Cazzamali	1937	M	45	Yes	Section	1 year 6 months	Yes	Yes	?	No	?	?

TABLE I.—(*Cont'd*) *Sacro-coccygeal Chordomas.* Cases Collected from the Literature

No.	Author	Year	Sex	Age	History of Trauma	Histol. Diagn. Section-Aspiration	Duration of Symptoms	Treatment		Recurrences	Metastasis	Survival Period	End Result
								Radiation	Surgery				
106.	Barnes & Owen	1937	M	59	No	Section	1 year	Yes	No	Yes	No	1 year 6 months	Dead of disease
107.	Bruce & Mekie	1937	F	44	Yes	Section	1 year	No	No	...	No	?	?
108.	Bruce & Mekie	1937	F	62	No	Section	1 year 6 months	Yes	Yes	Yes	No	?	?
109.	Odasso	1937	M	61	Yes	Section	1 year 6 months	No	Yes	Yes	No		?
110.	Bobbio	1938	M	54	No	Section	10 years	No	Yes	Yes	No	4 years	?
111.	Ghaleeb	1938	M	45	No	Section	2 years	Yes	Yes	Yes	No	2 years	Dead of disease
112.	Ghareeb	1938	M	55	No	Section	?	No	Yes	?	No	?	?
113.	Lyall	1939	F	42	No	Section	10 months	No	Yes	?	No	?	?
114.	Ashour	1939	M	50	Yes	Section	3 years	No	Yes	?	No	?	?
115.	Bowers	1940	M	61	No	Section	1 year	No	Yes	?	No	?	?
116.	Mignone	1940	M	68	No	Section	?	?	?	?	Lymph nodes Heart, lungs	?	?
117.	Mignone	1940	M	48	No	Section	?	?	?	?	No	?	?
118.	Kilby	1941	M	60	No	Section	3 years	Yes	No	Yes	No	2 years	?
119.	Harvey & Dawson	1941	M	63	Yes	Section	3 years	No	Yes	Yes	No	7 years	Living with disease
120.	Harvey & Dawson	1941	F	41	No	Section	2 years	No	Yes	No	No	3 years 6 months	?
121.	Harvey & Dawson	1941	M	67	No	Section	?	Yes	No	Yes	No	3 years	?
122.	Harvey & Dawson	1941	F	29	No	Section	4 years	Yes	Yes	Yes	No	3 years	?
123.	Olsman & Lev	1943	M	73	Yes	Section	5 years	No	Yes	Yes	No	3 years 6 months	Dead of disease
124.	Rehausen, et al	1943	F	25	Yes	Section	3 years	Yes	Yes	?	No	?	?
125.	Faust, et al	1944	M	23	No	Section	8 months	Yes	No	Yes	Lungs, adrenal muscle	1 year	Dead of disease
126.	Keane	1944	M	70	No	Section	4 years	Yes	Yes	Yes	No	1 year	Living with disease
127.	Costa & Filho	1944	F	53	No	Section	20 years	No	Yes	Yes	No	?	?
128.	Graf	1944	M	58	No	Section	?	Yes	No	Yes	Lymph nodes, liver, lungs, skin	4 years	Dead of disease

TABLE II.—*Sacro-Coccygeal Chordoma.* Memor

No.	Name	Age	Sex	Trau-ma	Bone Involve-ment	X-ray Diagnosis	Histol. Diagn. Section-Aspir.	Duration of Symptoms	Physical Examination	Treatment
1	A. K.	62	M	No	Distal ⅗ of sacrum	Chordoma	Asp. biopsy Chordoma	2½ yrs.	Large palpable tumor involving lower sacrum. Difficulty in starting urinary stream. Pain on the right leg.	*X-ray* (pre-op. 1500rx5 Surg.—wide ex sion of tumor *X-ray*—1400rx *X-ray*—900r (10cm cone)
2	M. McP.	50	F	Yes	Entire sacrum	Osteogenic sarcoma	Section Chordoma	3½ yrs.	Semi-fluctuant tumor over upper sacrum posteriorly. Urinary incontinence. Involuntary bowel movements. Partial anesthesia of rectum. Difficulty on walking.	*X-ray therapy* where (total unknown) *Radium Pack* 2800 mg. fo 2 ports
3	R. G. J.	70	M	No	3rd and 4th sacral segments	Chordoma	Asp. biopsy Chordoma	9 mos.	Irregular elastic tumor over left sacrum and the left pelvis posteriorly. Loss of sensation on sole, left foot. Knee jerk and ankle jerk decreased on the left side.	*X-ray:* 1600rx 2400rx *1 year later:* 400rx3 *5 months later:* 400rx7 (pelvic cycle) *6 months later:* Pelvic cycle re peated. *7 months later:* vic cycle repea
4	P. K.	60	M	No	Right side—lower 3 segments of sacrum. Coccyx also involved.	?Chordoma ?Giant cell tumor	Asp. biopsy Chordoma	2 yrs.	Round, well circumscribed cystic fluctuant mass overlying lower half of sacrum. Diminished sensation over saddle area on left. Motor weakness present. Knee jerk and ankle jerk diminished, particularly on the left.	*X-ray:* 1600rx 1 yr.-3 mos. la 1200rx3 *1 yr. later:* 1200rx2 *6 mos. later:* 400rx5
5	J. M.	37	M	Yes	Rt. side of the lower half of sacrum.	Area of bone destruction in the distal sacrum.	Section Chordoma	1 yr. 2 mos.	Smooth, soft, non-tender mass which extends upward for at least 8 cm. It is fixed to sacrum. No neurological signs, no nerves or roots involved.	*Surg:* Excision coccyx; partial cision of sac and curettage maining tu from the sa canal.
6	S. M.	62	M	No	Left side of the lower ⅗ of sacrum	Chordoma	Section Chordoma	Over 2½ yrs.	Large fixed 18 cm. mass occupying entire left buttock and anal cleft. Entire left side of pelvis filled with the mass. Pt. limps on walking, no urinary or rectal incontinence.	*X-ray:* 2800rx
7	C. T.	62	F	No	Lower end of sacrum and proximal segment of coccyx.	Suggestive of neoplasm with bone destruction	Asp. biopsy Chordoma	3 mos.	Hard mass measuring 25 cm. and extending from the sacroiliac region to the right greater trochanter. No neurological signs.	*X-ray therapy:* 1500rx2 *3 mos. later:* 8 *9 mos. later:* Removal of tu and sacrum.

I.—*Sacro-Coccygeal Chordoma.* Memorial Hospital Series

n ns	Physical Examination	Treatment	Clinical Course	Surviva Period
	Large palpable tumor involving lower sacrum. Difficulty in starting urinary stream. Pain on the right leg.	*X-ray* (pre-op.) 1500rx5 Surg.—wide excision of tumor *X-ray*—1400rx2 *X-ray*—900r (10cm cone)	Urinary incontinence following operation. Recurrence 2 years after operation.	5 yrs.
	Semi-fluctuant tumor over upper sacrum posteriorly. Urinary incontinence. Involuntary bowel movements. Partial anesthesia of rectum. Difficulty on walking.	*X-ray therapy* elsewhere (total dose unknown) *Radium Pack* 2800 mg. for 2 ports	Temporary improvement following the treatment by radio element pack.	11 mos.
	Irregular elastic tumor over left sacrum and the left pelvis posteriorly. Loss of sensation on sole, left foot. Knee jerk and ankle jerk decreased on the left side.	*X-ray:* 1600rx2 2400rx2 *1 year later:* 400rx3 *5 months later:* 400rx7 (pelvic cycle) *6 months later:* Pelvic cycle repeated. *7 months later:* Pelvic cycle repeated.	Fairly marked improvement following radiation. Intermittent recurrences of pain with remission after X-ray therapy. Lately pt. has had alcohol injections (4th lumbar interspace) for pain.	5 yrs. 4 mos.
	Round, well circumscribed cystic fluctuant mass overlying lower half of sacrum. Diminished sensation over saddle area on left. Motor weakness present. Knee jerk and ankle jerk diminished, particularly on the left.	*X-ray:* 1600rx3 1 yr.-3 mos. later 1200rx3 *1 yr. later:* 1200rx2 *6 mos. later:* 400rx5	Pt. improved following radiation. However, last one made him worse; severe pain, involving S-1 and S-5, rectal and bladder sphincters partially involved. Cordotomy performed; patient relieved.	4 yrs.
	Smooth, soft, non-tender mass which extends upward for at least 8 cm. It is fixed to sacrum. No neurological signs, no nerves or roots involved.	*Surg:* Excision of coccyx; partial excision of sacrum and curettage of remaining tumor from the sacral canal.	Free of disease until June when tumor recurred. Now has severe pain. Cordotomy performed and pt. completely relieved.	4 yrs.
	Large fixed 18 cm. mass occupying entire left buttock and anal cleft. Entire left side of pelvis filled with the mass. Pt. limps on walking, no urinary or rectal incontinence.	*X-ray:* 2800rx2	No improvement at all. No decrease in size of tumor. Developed metastasis—lower lobe of the right lung.	1 yr. 2 mos.
	Hard mass measuring 25 cm. and extending from the sacroiliac region to the right greater trochanter. No neurological signs.	*X-ray therapy:* 1500rx2 *3 mos. later:* 800rx2 *9 mos. later:* Removal of tumor and sacrum.	Uneventful postoperatively. Healing per primum. No symptoms.	1 yr. 4 mos.

	Survival Period	Autopsy	End Result	Comment
ce n. s	5 yrs.		Living with disease	Pt. had radiation preoperatively. After recurrence the patient has been kept fairly comfortable with radiation.
e- ie o	11 mos.	Retroperitoneal tumor attached to the sacrum; size of a baby head. Iliac bones and sacral plexus also involved.	Dead of disease	
a- g t- of n y. l- h e)	5 yrs. 4 mos.		Living with disease.	Surgery was feasible in this case, however, due to the patient's age and heart condition, was not carried out.
y- r- le re -1 d rs d. ied;	4 yrs.		Living with disease	In retrospect we believe this patient should have had an attempt at removal of the tumor.
il e- e- my ot. l.	4 yrs.		Living with disease.	Pt. was free of disease during 3 years and 6 months.
at in el- ow- ht	1 yr. 2 mos.		Dead of disease.	Pt. had been operated upon elsewhere twice and in both instances the tumor recurred.
er- er oms.	1 yr. 4 mos.		Dead of other cause. No evidence of disease.	Pt. died of leukemia and there was no clinical evidence of disease at time of death.

embryonal development a thickening of the entoderm occurs in a mid-sagittal plane known as the chordal plate and it has been shown that this plate becomes pinched off from the entoderm forming a longitudinal structure ventral to the neural canal—the primitive notochord (Fig. 1). The notochord then becomes surrounded by mesenchyme and these mesenchymal cells arrange themselves in segments, thus constituting the anlage of the vertebrae. Each of these

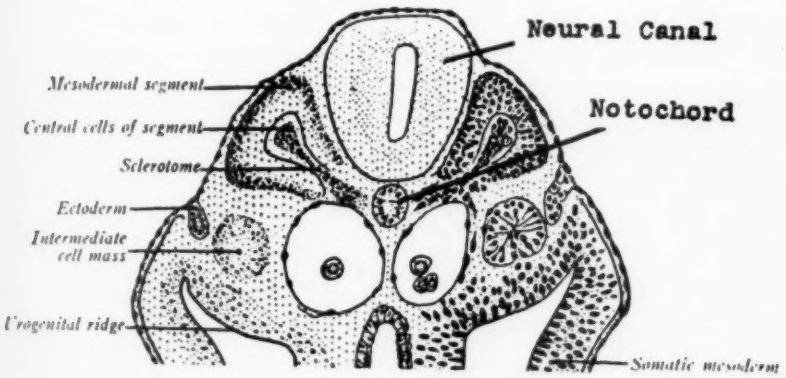


FIG. 1.—Transverse section of human embryo (Kollman).

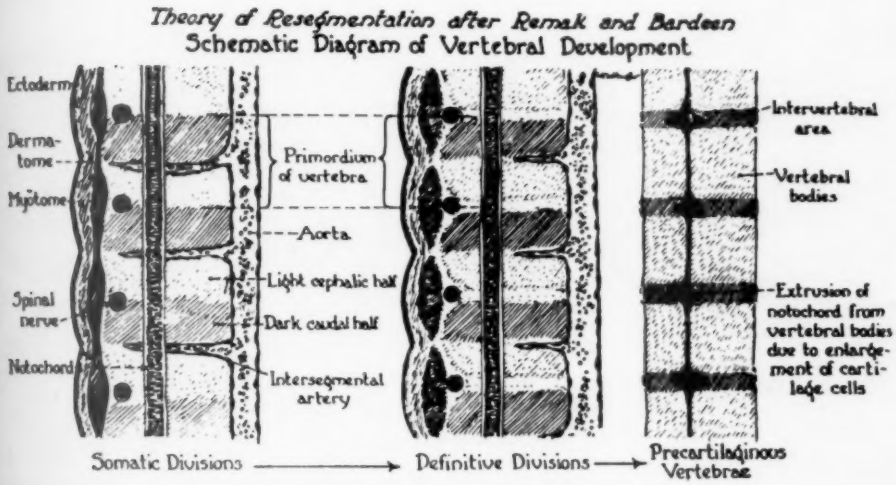


FIG. 2.—From Keyes and Compere.

sclerotomal segments surrounding the notochord is vascularized by an intersegmental artery. Between the segments there is an avascular area which remains undifferentiated for some time and this avascular zone represents the anlage of the intervertebral disc (Fig. 2).

In the embryo of ten weeks the vertebral body contains typical cartilage and ossification centers and at this time the tissue between the primitive vertebrae consists of elongated fibroblastic cells. As the intervertebral tissue is

compressed by the growing vertebral bodies, due to progressive cartilaginous formation and ossification, the cells on the periphery of the notochord are extruded so that the notochordal cells are only confined to the central portion of the intervertebral disc, i.e., the nucleus pulposus.

The bulk of available evidence indicates that chordomas arise from the primitive fibroblastic cells of the notochord. Although the present study is confined to an analysis of chordoma in the sacrococcygeal region, distribution of the tumor elsewhere along the vertebral column is of interest in this study, especially from the point of view of comparative anatomic incidence. Of the 161 reported cases of chordoma of all types encountered in the literature up to 1935 and including the Memorial Hospital series, 60 per cent were of the

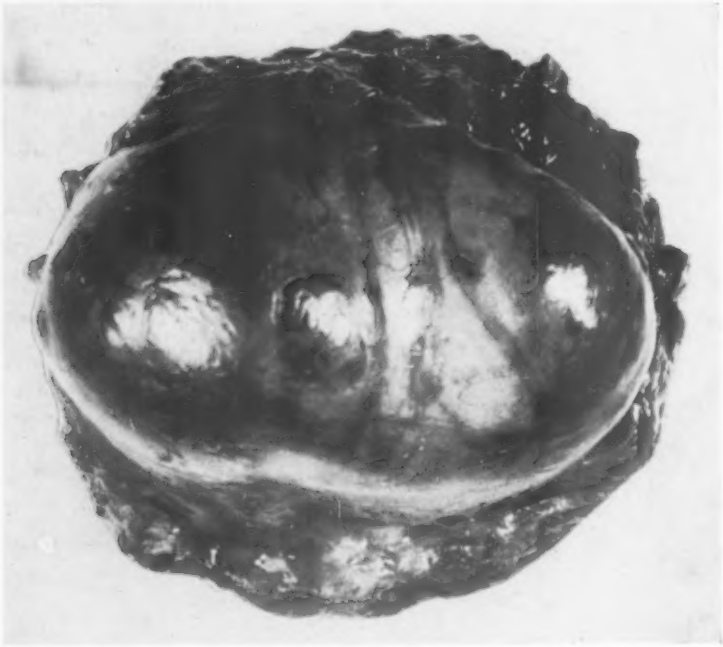


FIG. 3.—J. M. (Case 5): Surgical specimen of a lobulated and well encapsulated notochordal tumor.

sacrococcygeal type and about 30 per cent occurred in the spheno-occipital synchondrosis. The remaining 10 per cent were evenly distributed in the cervical, lumbar and thoracic regions. The predilection of chordoma for the sacrococcygeal area has never been satisfactorily explained, although some authors attribute this regional selectivity to the high incidence of trauma to the lower end of the spine. We have already mentioned the difficulty of evaluating the relationship of trauma to this tumor, or indeed to neoplasms in general.

PATHOLOGY

Gross Pathology. A chordoma is almost always not a resectable tumor even when only moderately advanced, hence few specimens are available for

gross morphologic study. In those instances where a reasonably wide surgical excision was performed (Fig. 3), the tumors were found to be bulky and encapsulated. The growth is characteristically lobulated and cystic in places, although true cyst formation does not occur. The surface of the tumor is purplish-red in color due to its excessive vascularity. On cut section, the neoplasm is seen to be composed of homogenous and translucent tissue in addition to irregular cavities filled with copious amounts of thin mucin.

Histology. The microscopic appearance of chordoma is characteristic in the average case and consists of large cells resembling bladder epithelium

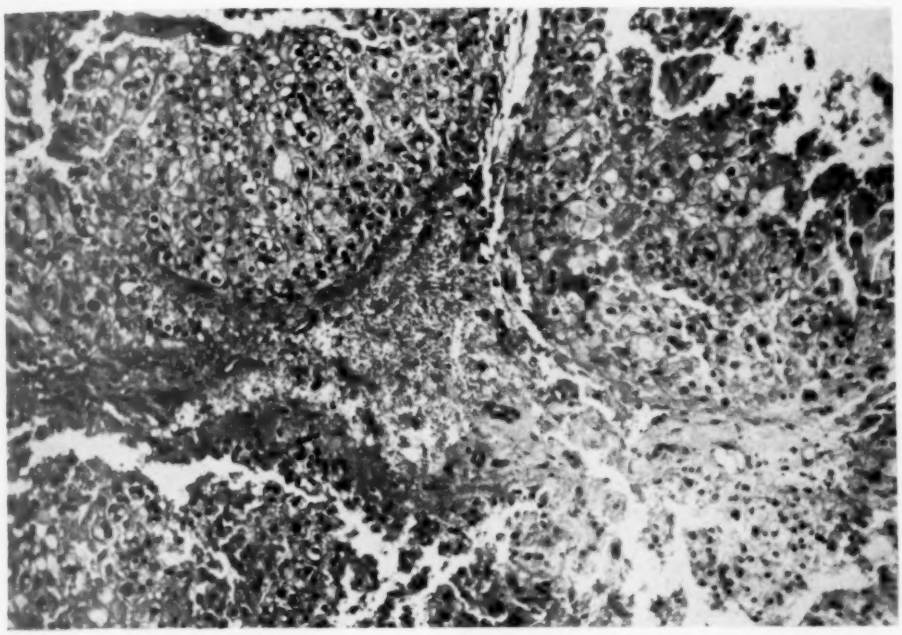


FIG. 4.—R. G. J. (Case 3): Aspiration biopsy (low magnification) showing the three main microscopic features, *i.e.*, presence of intra- and extra-cellular mucin, physaliphorus cells and lobular arrangement of the tumor cells.

(physaliphorus cells) which contain intracellular and extracellular mucin (a primitive function of notochordal tissue). These cells are arranged in lobules or cords, or they may present a solid epithelial pattern. In some instances the demarcation of the individual cells is lost and the microscopic appearance is that of a highly vacuolated syncytium. The presence of mucin and vacuolization together with the specific cells found in these tumors are the most important histologic features of chordoma (Figures 4 and 5). In the more malignant types, mitotic figures, cellular pleomorphism, hyperchromatic nuclei, and multi-nucleated giant cells are seen. Intracellular glycogen has been demonstrated in chordoma by some observers and this substance is probably responsible for the cytoplasmic vacuolization; nuclear vacuolization, however, is seldom seen.

Although this tumor is well differentiated and of relatively low grade malignancy, less differentiated and more highly malignant types are occasionally encountered, probably accounting for the widepsread metastases which occur in about 10 per cent of the cases.

SYMPTOMS, MORBID ANATOMY, AND CLINICAL COURSE

A chordoma is a slow-growing tumor and does not produce clinical manifestations until it is moderately well advanced. Our review of 128 cases of sacrococcygeal chordoma reported in the literature reveals that the average duration of symptoms was 20 months before the patient finally sought relief, and in two instances 20 years elapsed from the time of onset of symptoms until the patient consulted a physician (Costa and Filho[23]; and Reynes[77]).

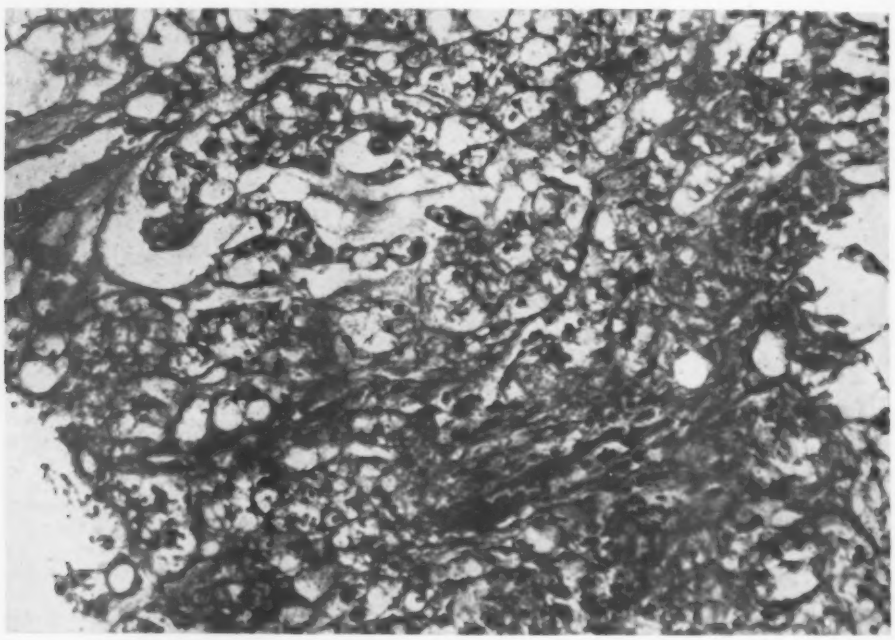


FIG. 5.—P. K. (Case 4): Aspiration biopsy (high power)—physaliphorus cells and intra- and extra-cellular mucin are seen.

Symptoms are directly dependent on the location of the growth and are due to expansion of a bulky neoplasm and to destruction of bone.

In sacrococcygeal chordomas pain is the earliest and most frequent symptom. It is generally mild and intermittent at first, becoming severe and intractable when the tumor involves the nerve roots and sacrum. In one-half of the cases, pain is present for many months before a tumor mass is noted. The patient's first complaint is usually pain in the rectal and anal regions, which frequently prompts the examining physician to make a diagnosis of hemorrhoids. Subsequent hemorrhoidectomy, of course, affords no relief. As the tumor begins to invade the rectum and urinary bladder, obstinate consti-

pation and urinary difficulties occur, and fecal and urinary incontinence complicate the clinical picture in most of the advanced cases. With involvement of the posterior nerve roots, motor and sensory disturbances of the lower extremities, gluteal region and external genitalia occur as well as trophic ulcers of the foot. In most of the cases the clinical course is slow but progressive, with eventual involvement of adjacent structures, particularly the sacrum. Destruction of the sacrum is an almost constant clinical manifestation of chordoma and is responsible for most of the morbidity produced during the early and moderately advanced stages of this disease. It was present in all of our cases and in one patient was complete (Case 2).

In the far-advanced cases, with widespread bone destruction involving the posterior nerve roots and pelvic vicera, the patient will present a picture of intractable pain, paraplegia and fecal and urinary incontinence. Intestinal obstruction and rectal and bladder hemorrhage may occur in the final stages of the disease.

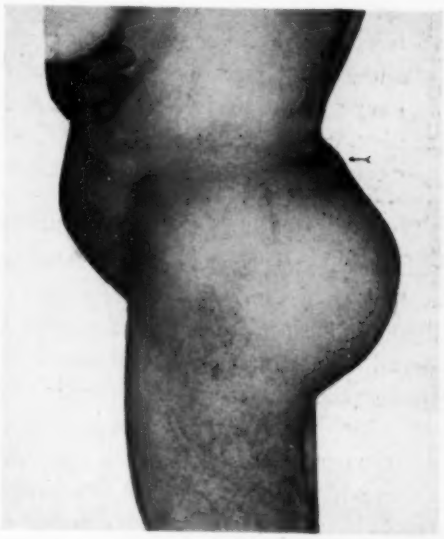


FIG. 6.—S. M. (Case 6): Clinical photo showing the tumor protruding in the lumbosacral region.

On the other hand there have been a few reported cases in which the course of the disease was fulminating. In 1933 Montgomery and Wolman[63] reported two cases of sacrococcygeal chordoma in which the duration of symptoms was only one month, followed by death from widespread metastases shortly thereafter. In one of our patients (Case 7) symptoms were present for only three months, at which time a bulky, well advanced sacrococcygeal chordoma was discovered.

The most constant physical finding in sacrococcygeal chordoma is the presence of a mass, the location of which will depend upon the *direction* of growth. If it is located anteriorly and protrudes into the pelvis, pushing forward the posterior wall of the rectum, no apparent tumor will be seen on inspection and external palpation, and only by digital rectal examination can such a growth be suspected. The hollow of the sacrum should always be carefully palpated, since a sacral tumor may easily be overlooked if only the anterior wall of the rectum is examined. Obviously if the direction of the growth is posterior, a visible and palpable external tumor is present (Fig. 6). Occasionally growth will extend in both directions (anteriorly and posteriorly) with production of both a pelvic and a postsacral mass. In the present series, anterior growth was present in three cases, posterior growth in two cases, and anterior and posterior growth in two cases; these proportions are in accord-

ance with observations of Stewart[87] and others. If the direction of growth is anterior, a pelvic mass will ultimately be present and in the late stages of this anatomic type of the disease, an intra-abdominal tumor will be noted. Regardless of the pattern of growth, palpation of the tumor will usually reveal it to be of firm consistency, though occasionally cystic areas are also present.

Curiously enough, the soft somatic tissues of the buttock and sacral region and the overlying skin are seldom involved by tumor, and cutaneous ulceration does not occur unless injudicious roentgen radiation has been given or surgical excision attempted followed by faulty wound healing. This can be ascribed to the fact that a chordoma is a slow-growing, expanding tumor and only becomes invasive and destructive when it meets with some resistance in the path of its growth as in the sacrum. The skin of the buttock is elastic and resilient so that in cases of massive, expanding tumor it can be pushed forward to a considerable degree without actually becoming involved by the disease.

Widespread metastases do occur, though rarely, particularly to the lungs and liver and the peripheral lymph nodes (Table III). Pulmonary metastasis was observed in one of our cases (Case 6) and was reported in approximately 5 per cent of the cases in the literature.

DIAGNOSIS

Since a sacrococcygeal chordoma produces a bulky mass externally or in the hollow of the sacrum, the condition should be suspected even though it might often simulate other neoplasms which occur in the sacrococcygeal area as well as non-neoplastic diseases in or about the pelvis. The tendency for chordomas to involve the pelvic viscera late in its course and the infrequency with which the overlying skin and subcutaneous tissues are involved, together with the occasional presence of cystic areas noted on physical examination are factors which should lead the examiner to suspect the possibility of a sacrococcygeal chordoma. As already emphasized, the sacrum is invaded and destroyed by a chordoma relatively early in its course so that roentgenographic studies will reveal some evidence of bone destruction in almost every case. These roentgenographic findings together with the clinical manifestations enumerated above warrant a tentative clinical diagnosis.

Differential Diagnosis. There are several conditions from which sacrococcygeal chordoma must be differentiated since they also may produce bulging, bulky tumefaction in the sacral and parasacral regions with or without bone destruction:

(1) Chondrosarcoma of the sacrum. Although this condition occurs but rarely, it may simulate a sacrococcygeal chordoma clinically because of the presence of a sacral tumor and roentgenographic evidence of bone destruction. However, chondrosarcoma of the sacrum does not usually produce a massive tumor until it is far advanced, at which time the surrounding soft tissues and skin are extensively involved and frequently there are associated pulmonary metastases. In the early stages of sacral bone sarcoma only roentgenographic studies will clearly differentiate the two conditions, and in the last analysis a

TABLE III.—*Sacro-Coccygeal Chordomas.* Cases which have metastasized

	Lymph nodes	Liver	Lung	Skin and Subcut. Tissue	Heart	Perito-neum	Muscle	Spleen	Kidney	Thyroid	Adrenal	Bladder	Omen-tum
1. Wagner (1910)	1	..	..	..	..	..	..	..	..	..	..	..	..
2. Pototsching (1919)	1	..	..	..	..	..	..	..	..	..	..	..	..
3. Peters (1919)	1	1	..	..	..	..	..	..	..	..	..	..	..
4. Lewis (1921)	1	1	..	..	..	..	..	..	..	..	..	..	..
5. Lewis (1921)	1	..	..	..	..	1	..	..	..	..	..	..	..
6. Lewis (1921)	1	..	..	..	..	..	..	..	..	..	..	..	..
7. Stewart (1922)	..	..	..	1	..	..	..	..	..	..	..	..	..
8. Arguad (1926)	..	1	..	..	..	..	..	..	..	..	..	..	..
9. Conway (1929)	1	..	..	..	..	1	1	..	..	..	..	1	1
10. Willis (1930)	..	1	1	1	1	..	..	1	1	1	..	..	..
11. Cato (1933)	..	1	..	..	..	..	..	..	..	..	..	..	..
12. Montgomery & Wolman (1933)	..	..	1	..	..	..	..	..	..	..	..	..	..
13. Mignone (1940)	1	..	1	..	1	..	..	..	..	..	..	..	..
14. Faust, et al (1944)	..	..	1	..	..	..	1	..	..	..	1	..	..
15. Graf (1944)	1	1	1	1	..	..	..	..	..	..	..	..	..
16. Memorial Hospital (1946)	..	..	1	..	..	..	..	..	..	..	..	..	..
	9	6	6	3	2	2	2	1	1	1	1	1	1

positive diagnosis can only be established by microscopic examination of a biopsy specimen.

(2) Tuberculosis of sacrum. Tuberculous infection of the sacrum is admittedly a rare occurrence but can produce bone changes together with abscess formation, resulting in a parasacral mass. When osseous tuberculosis advances to a stage where a bulky mass is produced, soft tissue infection (cold abscess) is invariably present and the clinical differential diagnosis should not be difficult. Evidence of tuberculosis elsewhere, together with positive bacteriologic findings, will aid in establishing a definite diagnosis of sacral tuberculosis.

(3) Tumors of the female pelvic organs. When a sacrococcygeal chordoma expands anteriorly to push the pelvic viscera forward, a pelvic, or occasionally an abdominal, tumor will be present, thus simulating the clinical picture of a neoplasm of the female genital tract. In one such case reported by Reich and Nechton,[76] a cystic pelvic chordoma clinically resembled an ovarian cyst in every way and the patient was even subjected to a laparotomy. Again, roentgen-ray studies of the sacral bone will almost always eliminate a primary tumor of the female genital tract since the latter does not usually invade bony structures.

(4) Tumors of the spinal cord. When the posterior nerve roots become involved by a chordoma, neurological manifestations appear in the lower limbs and the later course of the disease is characterized by fecal and urinary incontinence. Since these symptoms may also be produced by tumors of the spinal cord, the latter condition must be considered in the differential diagnosis. The differential diagnosis should not be difficult since neurological symptoms and signs occur early in lesions of the spinal cord, whereas bulky tumors in the sacrococcygeal region and pelvis are rarely encountered under these conditions.

(5) Tumors of the sacral soft parts. Benign and malignant tumors of the soft parts about the sacrum may clinically resemble sacrococcygeal chordoma in every way, especially when they assume massive proportions (neurofibroma,, neurogenic sarcoma, spindle cell sarcoma, etc.). Again, tumors of these soft parts seldom involve bone and then only late in their course, so that roentgenographic study of the sacrum is of considerable value in establishing the diagnosis. The final decision however will depend on the microscopic study of biopsy material.

(6) Sacrococcygeal teratoma. This rare and unique tumor is almost always encountered in infants and children and in most cases produces an external pedunculated growth without evidence of actual involvement of the sacrum.

(7) Cancer of the rectum. A colloid rectal carcinoma situated on the posterior rectal wall may often resemble sacrococcygeal chordoma which has expanded anteriorly to invade the rectal wall. In advanced cancers of this type with invasion of the pararectal tissues, the clinical differentiation may be difficult. Since this neoplasm, however, shows little tendency to bone involve-

ment, roentgen-ray studies of the pelvis are of considerable aid in distinguishing between the two conditions.

Roentgenographic Features. Roentgen-ray examination is an invaluable diagnostic aid since a sacrococcygeal chordoma involves the sacrum relatively early in its course in almost every case. Our analysis of previously reported cases shows that a correct radiographic diagnosis was made in only about 10 per cent of the cases, and in the Memorial Hospital series of seven cases the diagnosis of sacrococcygeal chordoma was made in four. In three of our cases where the correct diagnosis was not made on roentgen-ray examination, the roentgenologist's report was osteogenic sarcoma, giant cell tumor, and bone destruction of undetermined origin, respectively. Hsieh and Hsieh[44] made extensive roentgenographic studies of chordoma and stated that there are four signs which are rather typical of this tumor—(1) expansion (Fig. 7); (2) rarefaction or destruction (Figs. 8 and 9); (3) trabeculation; (4) calcification. We believe that if these roentgenographic manifestations are present in a case in which a bulky sacrococcygeal tumor is present, a diagnosis of chordoma is justifiable.

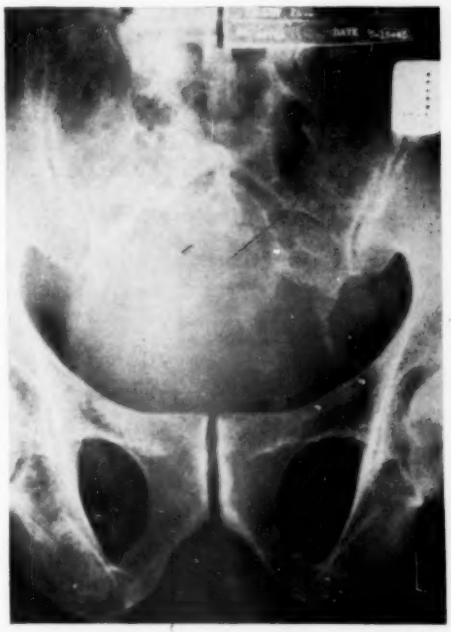


FIG. 7.—P. K. (Case 4): Roentgenogram of the pelvis showing bone *destruction* and *expansion*, which are characteristic of this growth.

Aspiration Biopsy. At the Memorial Hospital a final diagnosis is always dependent on a histologic examination of a tissue specimen wherever biopsy is possible. Aspiration biopsy has, in our experience, been a simple and effective method of establishing a microscopic diagnosis of chordoma. This method of tissue examination obviously requires the cooperation of a pathologist who is thoroughly familiar with and trained in the interpretation of aspirated tissue. Aspiration biopsy was performed in six of the seven cases in the present series and an unequivocal diagnosis of chordoma was made in four of these. An incorrect diagnosis of chondrosarcoma was made in one of the other two cases and in the last case the amount of tissue obtained by aspiration was insufficient for microscopic study. If a diagnosis cannot be established by aspiration biopsy, incisional biopsy will necessarily have to be done since no therapeutic regimen should be decided on without a definite anatomic diagnosis.

Of all previously reported cases in the literature, we could find only two where the diagnosis was made by aspiration biopsy (Micotti,[60] Richards, and

King[79]). If the technic of aspirating tissue for histological examination as described by Martin and Ellis[58] is followed and a competent tumor pathologist familiar with this material is available, aspiration biopsy should reveal the correct diagnosis in a high percentage of cases of chordoma, thus sparing the patient an incisional biopsy which may be followed by infection, ulceration or fungation.

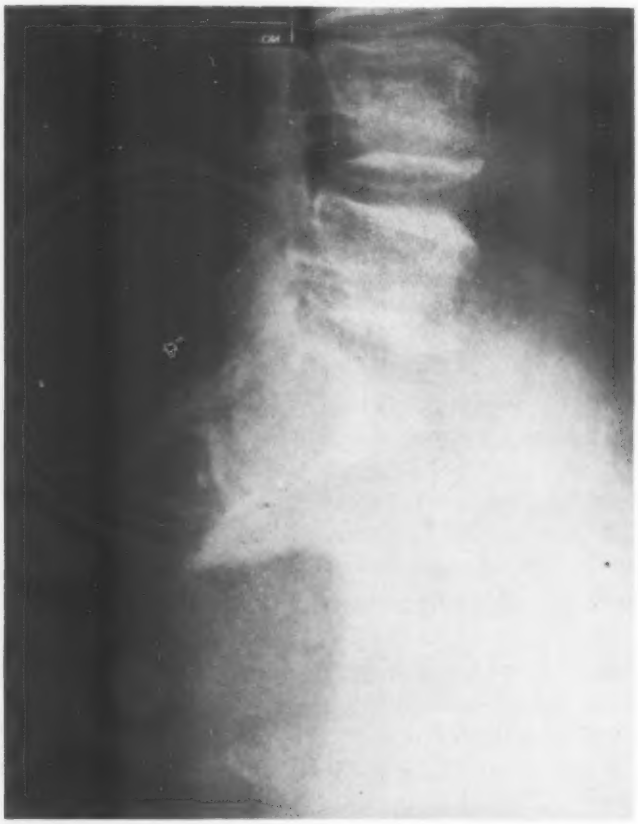


FIG. 8.—A. K. (Case 1): Lateral stereoscopic view illustrating *bone destruction*, one of the characteristic roentgenographic features of sacrococcygeal chordoma.

TREATMENT

It seems to be generally accepted by most authors that surgical treatment should be employed wherever possible and a survey of previously reported cases indicates that only in rare instances are sacrococcygeal chordomas even moderately radiosensitive. Since no appreciable regression of chordoma is obtained with radiation therapy and since complete extirpation of the tumor is not possible even when radical surgery is employed, the management of this neoplasm is chiefly a matter of palliation and partial control. It is theoretically possible, however, surgically to excise a small and early chordoma, but

this disease has seldom, if ever, been observed in its incipiency. The usual sequence of events in the treatment of sacrococcygeal chordoma is: repeated surgical excisions with or without radiation therapy followed by repeated local recurrences (6 to 18 months) over a period of years. Machulko and Rochlin[57] described a case of sacrococcygeal chordoma where the patient survived for 16 years during which period he was subjected to four surgical procedures combined with radiation therapy.

Although radiation therapy is of little value in most cases, it is reputed to be somewhat more effective in the treatment of chordoma in children. Montgomery and Wolman[63] cited three cases in children and in one of these satisfactory regression was supposed to have followed a course of roentgen radiation. Our experience with radiation therapy in the treatment of sacrococcygeal chordoma is that this mode of therapy does not induce regression, but is of some value in pain control, especially during the late stages of the disease. Repeated courses of roentgen radiation are not without danger, especially in this avascular area which is always subject to pressure and necrosis. Preoperative radiation therapy is contraindicated because surgery thereafter is likely to be complicated by failure of wound healing due to interference with the blood supply of the overlying soft tissue.

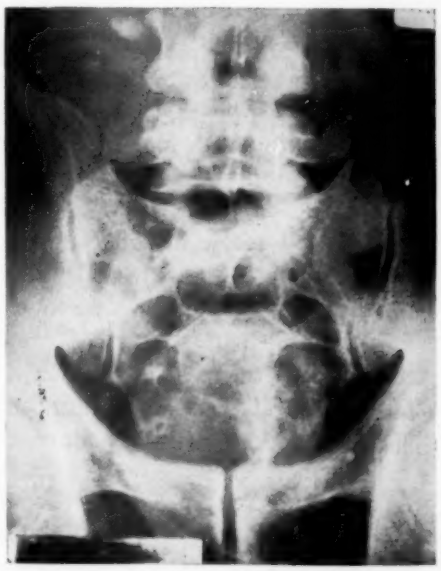


FIG. 9.—J. M. (Case 5): Roentgenogram of pelvis showing *bone destruction* of the lower half of the right side of the sacrum.

These cases nearly always reach a period when control of constant pain is the major problem and one difficult to meet. A spinothalamic chordotomy was employed in two of our cases with satisfactory results (Cases 4 and 5) insofar as pain relief was concerned. More frequent use of this procedure before it becomes necessary to administer opiates will be found to alleviate materially the unhappy lot of these patients. Chordotomy for chordoma should, of course, always be bilateral.

Until an operative technic is developed which envisages complete removal of the tumor-bearing segment of the sacrum and coccyx, the present unsatisfactory results of surgery will probably not be materially improved. Such radical surgery naturally involves difficulty with regard to bladder and rectal control. It has occurred to us that these complications might be overcome by preliminary permanent colostomy and uretero-enterostomy, yet to propose such major and objectionable procedures in the early stages of this disease

(when limited extirpation might be successful) may well be considered unduly radical by the surgeon and unacceptable to the patient. These limitations only serve to emphasize the extent of the difficulties presented by this disease.

It is nevertheless our opinion that surgical excision should be attempted whenever a recurrence occurs even though there is little likelihood of complete removal. Those cases cited in the literature in which there were long term survivals were essentially treated by repeated surgical excisions.

PROGNOSIS

Recurrence following surgical excision generally occurs from six months to within a few years, as already stated. Since sacral chordomas are characteristically slow-growing, numerous recurrences will occur over a period of many years following repeated excisions with massive invasion of the pelvis in the late stages of the disease. Occasionally, however, a single surgical excision will result in satisfactory control of the disease. Fletcher, *et al.*[32] reported a case of sacrococcygeal chordoma which had survived for seven years following operation without recurrence or metastasis. One of Seneque and Grinda's[83] patients survived seven years with no evidence of recurrence. Of 128 cases reported in the literature, regional and peripheral lymph node and other metastases occurred in only 15 patients (11 per cent) as shown in Table III. In more than 50 per cent of the metastatic cases, the most frequent sites of metastasis were the regional and peripheral lymph nodes, lungs, liver, and skin, respectively. The peritoneum, heart, and striated muscles were the sites of disseminated disease in two instances. Metastasis from sacrococcygeal chordoma has also been observed in the spleen, kidney, thyroid and adrenal glands, urinary bladder and omentum. In striking contrast to the sacrococcygeal chordoma, the spheno-occipital type almost never metastasizes and there has been only one such instance cited in the literature. Pulmonary metastases were demonstrated roentgenographically in one patient (Case 6) in the series herein reported.

END RESULTS

One of our patients (Case 5) has been treated by surgery alone and was free of disease for three years, after which he developed local recurrence. This patient had subsequently been submitted to a partial removal of the recurrent disease combined with a bilateral chordotomy and at the present is living comfortably four years after his first admission. In two other cases (Case 1 and 7) surgery was combined with roentgen therapy; one of the patients expired 16 months later of leukemia at which time there was no clinical evidence of either recurrent or metastatic disease. The other patient (Case 1) has lived, with recurrent disease, for five years. Radiation therapy alone was administered in four of our cases because the neoplastic process was either too far advanced or the patient's general condition was unsuitable for surgical treatment. Of these patients only two (Cases 3 and 4) are alive at present (five and four years, respectively) with recurrent disease. The other two patients

(Cases 2 and 6) have died of advanced recurrent disease within approximately a year from the date of admission.

These clinical experiences indicate that a cure cannot be obtained in cases of sacrococcygeal chordoma since the neoplasm does not lend itself to complete surgical extirpation and the most that can be hoped for with our present methods is palliation.

CONCLUSIONS

From a survey of the published records of 128 cases of sacrococcygeal chordoma and of seven hitherto unpublished cases from the Memorial Hospital the following conclusions are drawn:

1. The condition fortunately is rare.

2. It occurs more frequently in males and appears chiefly in the age groups of from 40 to 60.

3. Aspiration biopsy is a useful and reliable method of establishing the diagnosis histologically.

4. Though generally considered to be benign by many authors, sacral chordoma metastasizes in approximately 10 per cent of cases and causes the death of the patient either directly or indirectly in nearly all instances.

5. It is of long duration, slow growing, yields but little to intensive roentgen therapy and is not amenable to complete surgical removal owing to its inaccessibility and extensive involvement of the pelvic organs and spinal cord. Local recurrence following surgical excision is a constant feature.

6. Pain is the most serious symptom, especially when the disease is advanced, and is difficult to control. Chordotomy seems to offer the best measure of relief and should be employed more frequently.

7. Satisfactory methods of treatment have not as yet been developed. A method allowing more radical surgical extirpation is needed. A radical surgical program adequate to encompass sacrococcygeal chordoma in the average case would involve immediate loss of bladder and rectal control. Preliminary colostomy and uretero-enterostomy are drastic measures which the surgeon is loathe to apply for a slow-growing and relatively low-grade malignant tumor.

8. The difficulties in the management of sacrococcygeal chordoma have been presented.

BIBLIOGRAPHY

[1] Albert, H.: Chordoma, with the report of a malignant case from the sacrococcygeal region. Surg. Gynec. & Obst., **21**: 767–770, 1915.

[2] Alexander, W. A., and J. W. Struthers: Sacro-coccygeal Chordoma. J. Path. and Bact., **29**: 61–64, 1926.

[3] Alezais and Peyron: Sur l'evolution cellulaire du tissu notochordal dans les tumeurs. Compt. rend. Soc. de Biol., Paris, **84**: 368, 1920.

[4] Andler, R.: Chordoma of Sacro-coccygeal Region. Arch. f. Klin.; Chir., **143**: 467–490, 1926.

[5] Argaud, R.: Chordome malin.; Arch. de Med. exper. et Anat. Path. Par., **28**: 364-374, 1918.
[6] Argaud, R.: Sur la phase carcinomatoid du chordome malin. Compt. rend Soc. de biol., Par., **82**: 428, 1919.
[7] Ashour, M. A.: Chordoma of Anterior Aspect of Larynx and Sacro-coccygeal Region; 2 cases. J. Egyptian M. A., **22**: 163–166, 1939.
[8] Barnes, V., and S. E. Owen: Case with unusual endocrine findings (prolan in urine). Am. J. Cancer, **29**: 541–545, March, 1937.
[9] Berard, Dunet, Peyron (1920): Bull. Assoc. fr. pour l'etude du cancer, Mai 1922.
[10] Bobbio, L.: Sacro-coccygeal Chordoma, clinical study of case. Boll. e mem. soc. piemontese di chir., **8**: 4–9, 1938.
[11] Bowers, W. F.: Sacro-coccygeal Chordoma, case. Nebraska M. J., **25**: 341–342, 1940.
[12] Bruce, J., and E. Mekie: Chordoma. Surg. Gynec. & Obst., **65**: 40–47, 1937.
[13] Bustos, F. M.: Case of Chordoma. Semana Med., **2**: 221–229, 1928.
[14] Cameron, J. A. M.: Case of Sacro-coccygeal Chordoma. Glasgow M. J., **106**: 38–43, 1926.
[15] Cato, E. T.: Australian & New Zealand J. Surg., **1**: 425–430, 1932.
[16] Cazzamali, P.: Central Sacral Chordoma; Histogenetic, Anatomico Pathologic and Clinical Study of Case. Arch. Ital. di. chir., **45**: 175–199, 1937.
[17] Chesky, V. E.: Sacro-coccygeal Chordoma. Arch. Surg., **24**: 1061–1067, 1932.
[18] Coley, B. L.: Sacral Chordoma One Year After Radical Resection. Ann. Surg., **105**: 463–466, 1937.
[19] Comby, J.: Chordoma in Children with Report of Three Cases. Arch. de med. d. enf., **37**: 671–679, 1934.
[20] Conway, C. A. G.: Sacro-coccygeal Chordoma with Extensive Metastases. Mag London (Roy. Free Hosp.), School Med. for Women, **241**: 7–9, 1929.
[21] Cordovil, A.: Clinical and Anatomo Pathologic Study, Case of Sacro-coccygeal Chordoma. Rev. bras. de cir., **3**: 443–463, 1934.
[22] Coulthard, H. S., and R. I. Haris: Chordoma, three cases. Canad. M. A. J., **33**: 522–527, 1935.
[23] da Costa, D. G., and N. Dunham Filho: Lumbosacral Chordoma, case. Rev. bras. de. cir., **13**: 649–656, 1944.
[24] Daland, E. M.: Boston M. & S. J., **180**: 571, 1919.
[25] Dickson, J. A., and C. A. Lamb: Sacral Chordoma. Ann. Surg., **93**: 857–861, 1931.
[26] Donati, A., and L. Manginelli: Chordoma of Sacro-coccygeal Region. Folia clin. et biol., **5**: 45–55, 1933.
[27] Estella, J.: Aberrant and Regular Sacro-coccygeal Chordomas, 2 cases. Arch. Fac. de Med. de Zaragoza, **1**: 447–456, 1932.
[28] Ewing, James: Neoplastic Diseases.
[29] Fabricius-Moller: A Case of Malignant Chordoma with Involvement of the Pharynx. Hosp. Tid., Kobenh, **62**: 849–857, 1919.
[30] Faust, D. B., and H. R. Gilmore, Jr., and C. S. Mudgett: Review of Literature with Report of Sacro-coccygeal Case. Ann. Int. Med., **21**: 678–698, 1944.
[31] Fischer, B., and Steiner: Ueber ein Maligner Chondrom der Schadel-Ruckgratshohle. Bentr. z. path. Anat. u.x. allg. Path., Jena, **11**: 109–119, 1906.
[32] Fletcher, E. M., H. W. Woltman, and A. W. Adson: Sacro-coccygeal Chordomas, a Clinical and Pathological Study. Arch. Neurol. & Psychiat., **33**: 283, 1935.
[33] Fumegalli, C. R.: Chordoma of Sacro-coccygeal Region. Clin. Chir., **30**: 1123–1158, 1927.
[34] Gabriel, W. B.: Sacro-coccygeal Chordoma, case. Proc. Roy. Soc. Med., **29**: 1007-1008, 1936.
[35] Gerber, J.: Chordoma of Sacrum, case. Arch. f. Klin., Chir., **159**: 248–256, 1930.

[36] Ghareeb, A. A.: Chordoma, Two Sacro-coccygeal Cases. J. Egyptian M. A., **21**: 606–613, 1938.

[37] Graf, L.: Sacro-coccygeal Chordoma with Metastases. Arch. Path., **37**: 136–139, 1944.

[38] Grahl, O.: Eine Ecchondrosis physaliphora spheno-occipitalis (Chordom des Turkensattles) Ungewohnlichen Umgang mit interessanten klinischen Folgen. Inaug. Dissert., Gottingen, 1903.

[39] Grandclaude, C., J. Driesseus and P. Tison: Echo Med. du nord, **37**, 186–189, 1933.

[40] Harmos, O., and L. A. Palmer: Chordomata and Report of Case. Virginia Med. Monthly, **62**: 638–648, 1936.

[41] Harvey, W. F., and E. K. Dawson: Chordoma. Edinburgh M. J., **48**: 713–730, 1941.

[42] Henning, L.: Uber congenitale echte Sakvaltumoren. Beitr. z. path. Anat. u.z. allg. Path., **28**: 593–619, 1900.

[43] Hirsch, E. F., and M. Ingalls: Sacro-coccygeal Chordoma. J. A. M. A., **80**: 1369–1370, 1923 (illus.).

[44] Hsieh, C. K., and H. H. Hsieh: Roentgenologic Study of Sacro-coccygeal Chordoma. Radiology, **27**: 101–108, 1936.

[45] Hutton, A. J., and A. Young: Malignant Sacro-coccygeal Chordoma and Chordoma of Dorsal Spine. Surg. Gynec. and Obst., **48**: 333–344, 1929.

[46] Keane, W. E.: Chordoma, case. J. Michigan M. Soc., **43**: 232–233, 1944.

[47] Keyes, Donald C., and Edward L. Compere: The Normal and Pathological Physiology of the Nucleus Pulposus of the Intervertebral Disc. J. Bone & Joint Surg., **30**: 897, 1932.

[48] Kilby, W. L.: Chordoma, case. Bull. School Med. Univ. Maryland, **26**: 11–16, 1941.

[49] Kolmann, Julius: Hanatlas der entwicklungs geschichte des menschen. Berlin, **2**: 1907.

[50] Kwartin, B., and J. D. Stewart: Sacro-coccygeal Chordoma. Ann. Surg., **86**: 771–775, 1927.

[51] Letulle, M., and Dujarier: Chordoma in Anterior Sacro-coccygeal Region, case. Bull. de l'Assoc. franc. p. l'etude du cancer, **16**: 308–315, 1927.

[52] Lewis, N. D. C.: Contribution to Study of Tumor from Primitive Notochord. Arch. Int. Med., **28**: 434, 1921.

[53] Linck, A.: Chordoma malignum, ein Beitrag. zur kenntius der Gerchwulste an der Schadelbasis. Beitr. z. path. Anat. u.z. allg. Path., Jena, **46**: 573–585, 1909.

[54] Lund, F. B.: Chordoma and Sacro-coccygeal Dermoid. Boston M. & S. J., **181**: 704, 1919.

[55] Lyall, A.: Chordoma (case of sacro-coccygeal region). Glasgow M. J., **131**: 171–172, 1939.

[56] Mabrey, R. E.: Chordoma, Study of 150 Cases. Am. J. Cancer, **25**: 501–517, 1935.

[57] Machulko-Horbatzewitsch, G. S., and L. L. Rochlin: Clinical Study, pathomorphology and histogenesis of chordomas. Arch. f. Psychiat., **89**: 222–262, 1930.

[58] Martin, Hayes and E. B. Ellis: Am. J. Surg., **96**: 169, 1930.

[59] Mazzia, O.: Chordom der Sakralgegend. Centralbl. f. allg. Path. u. path. u. Anat., Jena, **21**: 769–772, 1910.

[60] Micotti, R.: Chordoma of Sacrum. Policlinico (sez. chir.), **29**: 265–281, 1922 (ills.); ab. J. A. M. A., **79**: 415, 1922.

[61] Monlonguet, P.: Sacro-coccygeal Chordoma. Rev. crit. du path. et de therap., **2**: 217–221, 1931.

[62] Monserrat, J. L., and M. L. Olascoaga: Sacro-coccygeal Chordoma in Childhood, with report of case. Rev. Assoc. Med. Arg., **51**: 106–113, 1937; also Prensa Med. Arg., **24**: 1889–1897, 1937.

[63] Montgomery, A. H., and I. J. Wolman: Sacro-coccygeal Chordomas in Children. Am. J. Dis. Child., **46**: 1263–1281, 1933.

[64] Muller, H.: Über das Vorkommen von Resten der Chorda dorsalis bei menschen nach

der Geburt und uber ihr Verhaltniss zu den Galbertgeschwülsten am Clivus Zeitschr. f. rat. med., **2**: 202–229, 1858.

65 Nash, I. E., and N. F. Laskey: Sacro-coccygeal Chordoma, report of unusual case with special reference to x-ray findings. J. Urol., **38**: 81–90, 1937.

66 Nebelthau, R. A.: Ueber die Gallertgeschwülste am Clivus Blumenbachii. Inaug. Diss., Marburg, 1897.

67 Odasso, A.: Sacro-coccygeal Chordoma; pathogenic and anatomico pathologic study of case. Arch. Ital. di chir., **37**: 505–531, 1934.

68 Olsman, L., and E. M. Lev: Sacro-coccygeal Chordoma, case. Am. J. Surg., **60**: 115–118, 1943.

69 Pai, H. C.: Sacro-coccygeal Chordoma, case. Chinese M. J., **46**: 456–464, 1932.

70 Penkert, M.: Early Stages of Ant-sacral Chordoma, 2 cases. Zentralbl. f. Gynak., **57**: 80–88, 1933.

71 Peters, W.: Ein rezidivierendes bostariges Chordom der Sacro-coccygealen Gegend mit metastasen. Deutsche Ztschr. f. Chir., Leipz., cli 191–199, 1919.

72 Podlaha, J., and F. Pavlica: Sacro-coccygeal Malignant Chordoma, case. Shorn. lek, **29**: 383–416, 1927.

73 Pototschnig, G.: Ein Fall von malignem. Chordom mit metastasen. Beitr. z. path. Anat. u.z. allg. Path., Jena, **65**: 356–362, 1919.

74 Pupo, M. D., Cardenas, and I. Calvo Tarafa: Histopathology of Chordoma of Sacro-coccygeal region. Arch. de med. int., **1**: 439–462, 1925.

75 Rahausen, A., A. Riesco, and I. Brieva: Chordoma with report of a case. Rev. Med. de Chile, **71**: 903–908, 1943.

76 Reich, W. J., and M. J. Nechtow: Cystic Pelvic Chordoma Simulating Ovarian Cyst. Am. J. Obst. & Gynec., **49**: 265–268, 1945.

77 Reynes, H., and Rouslacroix: Large Sacro-coccygeal Chordoma. Bull. de l'Assoc. Franc. p. l'etude du cancer, **16**: 434–446, 1927.

78 Ribbert: Uber die Ecchondrosis physaliphora spheno-occipitalis. Centrbl. f. allg. Path., **5**: 457–461, 1894.

79 Richards, V., and D. King: Chordoma. Surgery, **8**: 409–423, 1940.

80 Richardson, A., and A. L. Taylor: Case of Sacro-coccygeal Chordoma. Brit. M. J., **1**: 862–864, 1926.

81 Riche, V., P. Lonjon, Guibert and Quet: Voluminous Sacro-coccygeal Chordoma, case. Arch. Soc. d. sc. med. et biol. de Montpellier, **16**: 469–474, 1935.

82 Sandahl, C.: Chordoma malignum sacro-coccygeale, case. Acta, Chir. Scandinav., **72**: 421–430, 1932.

83 Seneque, J., and J. P. Grinda: Sacro-coccygeal chordoma, cases. J. de Chir., **35**: 817–842, 1930.

84 Sommer, L.: Case of Antesacral Heterotypical Chordoma. Med. Clin., **22**: 692–693, 1926.

85 Stanton, O. L.: Chordoma with Report of 2 Cases. Canad. M. A. J., **27**: 20–22, 1932.

86 Stewart, Fred W.: Occupational and posttraumatic cancer. Bull. New York Acad Med., March, 1947.

87 Stewart, M. J.: Malignant Sacro-coccygeal Chordoma. J. Path. & Bacteriol., **25**: 40–62, 1922 (illus.).

88 Stewart, M. J., and J. E. Morin: Chordoma, Review with Report of New Sacro-coccygeal Case. J. Path. & Bact., **29**: 41–60, 1926 (illus.).

89 Stewart, M. J., and E. Morin: Chordoma of Coccyx, diagnosis and treatment—88 cases. Bull. Med. de Quebec, **29**: 1377, 169, 1928.

90 von Tempsky, A.: Sacro-coccygeal Chordoma. Zentralbl. f. Chir., **56**: 2892–2894, 1929.

91 Tuffier, Gery L., and Vignes: Chordome malin de la region sacro-coccygienne. Bull. et Mem. Soc. Anat. de Chir., **89**: 459–469, 1914–1919.

92 Vecchi, A.: Recidivirendes bostartiges Chordom der sacro-coccigealen Gegend. Arch. f. Klin. Chir., **94**: 575–589, 1912.

93 Virchow: Entwicklung des Schadelgrundes; Die Krankhaften Geschwulste.

94 Wahling, F.: Malignant Sacro-coccygeal Chordoma, case. Med. Klin., **28**: 645–646, 1932.

95 Walz, K.: Case of Malignant Chordoma. Munchen. Med. Wochnschr., **72**: 1241–1242, 1945 (illus.).

96 Wildbolz, E.: Malignant Chordoma of Sacrum, case. Zentralbl. f. Chir., **58**: 2875–2880, 1931.

97 Willis, R. A.: Sacral Chordoma with Widespread Metastases. J. Path. & Bact., **33**: 1035–1043, 1930.

98 Zerbini, E. de Jesus: Sacro-coccygeal Chordomas. Rev. de cir. de São Paulo, **5**: 355–394, 1940.

PLASMA SILK SUTURE OF NERVES

JAMES E. BATEMAN, M.D., F.R.C.S.(C)
TORONTO, CANADA

FROM THE DIVISION OF ORTHOPAEDIC SURGERY, DEPARTMENT OF VETERANS AFFAIRS, CHRISTIE STREET HOSPITAL, TORONTO, ONTARIO

SURGEONS CLOSELY CONCERNED with the repair of injured nerves constantly strive to improve the technic of suture. No matter how many sutures have been done, the operator always feels that his last one could have been just a little better. Accuracy of alignment, adjustment of tension, control of rotation, atraumatic technic are criteria of good peripheral nerve surgery and constant challenges to the attainment of perfection. Of the technical suggestions for improvement which have appeared, that which comes closest to the ideal is some type of adhesive or glue neurorrhaphy.

DEVELOPMENT OF THE GLUE TECHNIC

There have been many attempts to introduce such a method dating back to Lotheissen in 1901 who used gelatin as the adhesive. Seddon and Associates made a most promising contribution during World War II in introducing the use of cockrel plasma fortified by tissue extract. This technic was not generally recommended because of undesirable foreign body reaction initiated by heterogeneous plasma and because of technical difficulties in application. It was further felt that the method of glue suture had no place where significant tension was present at the suture line. The introduction by Tarlov and Associates of autogenous plasma as an adhesive using a removable rubber mould placed the method on a more practical basis. The used of adhesive alone was still not feasible under even moderate tension. Spurling then suggested the use of tantalum sutures to support the clot. Experimental investigation of the method at this center resulted in our modifying the method of plasma preparation and replacing tantalum with silk. This plasma silk method has now been used in some 350 human nerve sutures. The views and conclusions in the ensuing discussion are based on the experimental and clinical application of the method.

DESCRIPTION OF PLASMA SILK METHOD

(1) *Preparation of Plasma.* Plasma suitable as an adhesive may be easily obtained from whole blood. The method suggested by Tarlov was investigated experimentally before application to human cases. An adequate yield of plasma is produced by centrifuging blood in sterile paraffin-lined iced test tubes without using an anticoagulant. Examination of early experimental sutures using plasma prepared by this method showed a foreign body reaction about fine granule-like areas in the plasma. The particles initiating this reaction were identified as flecks of paraffin, separated in the centrifuging process and which

TABLE I.—*Silk Sutures*

Nerve	No.	Motor Recovery	Sensory Recovery	Motor and Sensory Recovery	Some or Partial Recovery
Ulnar	11	4	7	4	7=64%
Radial	7	4	5	4	5=71%
Median	5	1	2	1	3=60%
Sciatic	10	7	2	2	7=70%
Peroneal	9	3	3	3	3=33⅓%
Total	42	19	19	14	25
Percentage	..	43%	43%	33%	59%

TABLE II.—*Plasma Silk Sutures*

Nerve	No.	Motor Recovery		Sensory Recovery	Motor and Sensory Recovery	Some or Partial Recovery
		Upper Group	Lower Group			
Ulnar	52	22 of 24	21 of 52	38	21=49%	59=96%
Radial	31	23 of 25	28 of 31	29	29=93%	29=93%
Median	24	10 of 12	14 of 24	20	14=58%	22=91%
Sciatic	19	13 of 19	6 of 19	8	8=44%	14=77%
Peroneal	15	10		12	10=67%	12=75%
Total	141	113		107	82	127
Percentage	..	80%		76%	62%	86%

become suspended in the plasma fraction. When the paraffin was removed from the technic, this reaction disappeared. In practice, 30 cc's. of blood are withdrawn from the patient into a sterile test tube after operation is started. This is centrifuged in ice-lined containers for three minutes at 2500 revolutions per minute. Some adjustment of time and speed of the centrifuge may be necessary because of variability in apparatus. When this is done, a standard yield is obtained. The plasma is kept in ice until needed and removed by a sterile pipette for application about the suture line. Proper cooling is the most important consideration in producing plasma without an anticoagulant, if attention is paid to this detail, the plasma will remain fluid and usable for 2–3 hours. Beyond this time, the tensile strength of the clot is diminished although the plasma may still be fluid.

(2) *Approximation of Nerve Ends.* Efforts have been made to completely avoid using any suture material but this has not been practical. Experimentally, sciatic nerves of dogs were united under moderate tension using a pull out suture after the fashion of Bunnell for tendon sutures which was removed

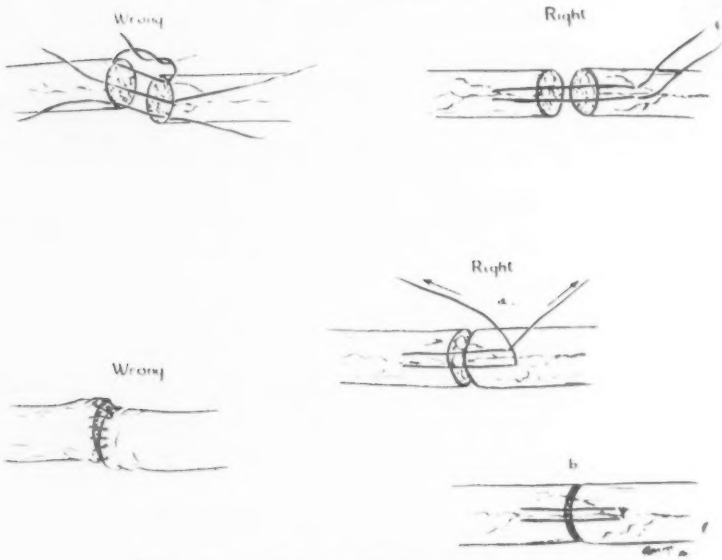


FIG. 1.—Insertion of the parallel stay sutures.

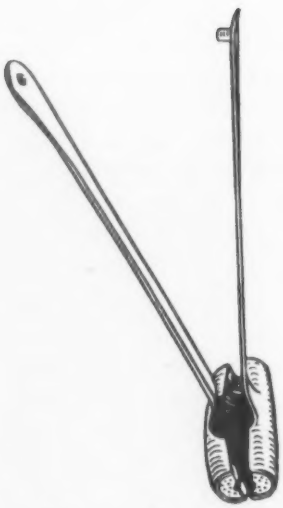


FIG. 2. — The spoon mould. Note the sponge rubber cuff at the ends.

after an autogenous clot was applied. Examination at the end of 48 hours showed the nerve ends to be completely separated. If an adhesive is to be used, it should be regarded rather as a supporting mechanism with tension largely controlled by a small number of sutures. In practice, it has been possible to reduce the number of sutures necessary to two of the perineural stay type when a clot is used in addition.

The sutures are inserted about ¼ inch from the prepared nerve ends and dip into the perineurium but not into subjacent fasciculi. They are applied one on each side of the trunk in parallel fashion (Fig. 1). The knots are tightened gradually so that the desired approximation and adjustment is obtained. This enables minute correction of the apposed surfaces.

(3) *Application of Clot.* The removable rubber mould or cup as suggested by Tarlov[4] has simplified this step considerably. In addition to this a combined mould and spoon with a cuff as illustrated has also (Fig. 2) been useful. After the stay sutures have been inserted, the cup is fixed in place and a final minute adjustment of the nerve faces is done through the opening provided. Bleeding from nerve ends must be controlled before the clot is applied. In this connection the rubber cuff serves as a hemostat as well as preventing leakage of the plasma. The cool plasma is removed in a sterile pipette or eye dropper, warmed gently by hot saline sponges and dropped into

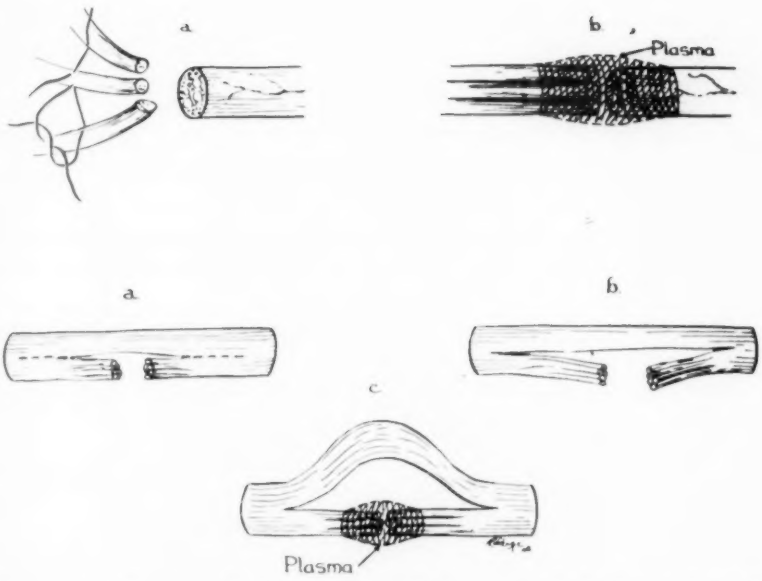


FIG. 3.—Application of plasma silk method to branch lesions and notch defects.

FIG. 4.—A satisfactory clot at operation.

the cup. The container is filled to the top and the plasma is allowed to clot. A satisfactory clot is obtained in 3–7 minutes, retraction (Fig. 4) is encouraged by gently separating the plasma from the edges of the cup. When firm, the clot is separated and the container gently removed.

COMPARATIVE CLINICAL RESULTS FOLLOWING SILK AND PLASMA SILK METHODS

In this study, a control series of routine silk sutures has been done and is compared with a series by the plasma silk technic so that some definite conclusions may be reached. There are many difficulties in setting up comparable groups of this nature for study. A most desirable common denominator is

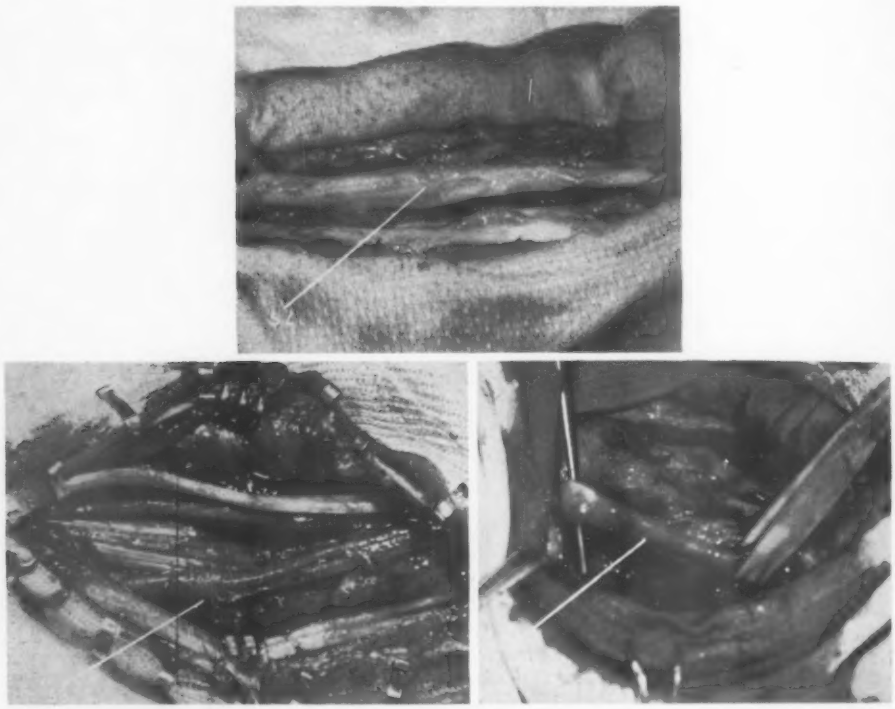


FIG. 5.—(A) Human plasma suture three months after operation. S.L. suture line.
(B) Human plasma suture four months after operation.
(C) Human plasma suture six months after operation.

that all the sutures be done by the same operator. This has been accomplished in this series. A completely analogous series would entail sutures at the same level, on the same nerve, in the same patient on opposite sides, which obviously is not possible. Sutures comparable as to site, length of time following injury, length of gap, severity of injury, *etc.*, have been reviewed. The figures presented comprise a portion only of the total number of operations. They include those cases in which recovery in part could be reasonably expected, estimating regeneration at the rate of 1 inch per month from the level of nerve union.

The silk sutures have been assessed at a longer interval following operation than has been the case with the glue sutures. The treatment apart from the method of nerve suture has been the same in all cases. The postoperative routine included monthly changes of plaster until the extremity was straight. Electrical stimulation was initiated through plaster as soon as the operative wound was healed and was continued as far as possible until voluntary power returned. The assessment of recovery in this series has been on motor and sensory return only. When even a carefully elicited Tinel test is done, apparent progress can be demonstrated in all cases. It is felt that any evaluation of results based on Tinel's sign alone is not reliable. Electromyographic studies of progress have also been done but for the present purposes of comparison, even this evidence has not been documented as definite recovery. The return of voluntary power has been regarded as the most reliable evidence of nerve regeneration but appreciation and accurate localization of touch, pinch and pin prick in the zone of autonomous supply have also been interpreted as recovery.

RESULTS

The following tables show in summary figures comparing plain silk and plasma silk technics.

ANALYSIS OF RESULTS

1. *Discussion of Comparative Figures.* The figures presented do not constitute a large enough series to be considered statistically. Recovery, both partial and complete has been superior in all groups sutured by the plasma silk technic. The difference is significant in all nerves but is most striking in the peroneal nerve and least in the case of the sciatic.

2. *Further Discussion.* (a) Limitations: The complete plasma silk routine could not be applied in all instances. Sutures in certain inaccessible regions made application of the cup awkward. For example, in tight sutures of the radial nerve at the elbow, acute flexion of forearm makes the insertion of the container difficult and pooling of the plasma only may be done. Despite extensive exposure and mobilization, some injuries were still encountered in which severe tension was present. These gaps sometimes need central transfixion sutures and multiple rim sutures as well so that any contribution by the plasma is at a minimum. However, improved control of infection and early secondary closure of wounds has decreased the incidence of irreparable gaps, paving the way for wider use of a meticulously accurate method.

3. *Advantages of Plasma Silk Method.* The explanation for the superiority of the combined technic is apparent in technical advances rather than in any magic contribution by the plasma to the process of regeneration.

(a) Control of Nerve Ends. The placing of the cut surfaces so that they face each other squarely without distortion is a delicate and fussy maneuver. In the classical method of multiple rim stitches, each new bite may sink to a different level, altering the control and direction of the preceding stitch. The parallel stay sutures provide a pivot affording minute correction and final adjustment of the ends before being solidified by the clot.

(b) Tension Adjustment. An equally difficult and frequently abandoned task is that of obtaining just the right amount of tension at the suture line. As each new suture is inserted, the fine adjustment is disturbed in tightening the knots. A further variable occurs since no two will be fixed with exactly the same tension. The usual land mark in multiple stitch insertion is the retracted epineurium. The edge may present at a different level at various points of the circumference so that it is an unreliable land mark. Unequal tension resulting from multiple sutures favours entanglement and buckling together of nerve bundles. In ends tightly apposed, compression inserts the proximal bundles between the distal tubes instead of leaving them in a relaxed facing position. As regeneration proceeds, angulation and confusion is increased, stifling smooth flow across the gap.

(c) Suture Material. Selection of the most desirable suture material involves several considerations. The structure of the substance controls the foreign body tissue reaction but an equally important consideration is the amount of that substance needed for a secure union. No advantage is gained in using a fine inert thread if double the amount is needed for the suture. The consistency of the suture material has a bearing on the security of the knot which is produced. The uneven surface of silk and similar substances has a resistance or friction which favours a secure grip in tying. Material such as wire on the other hand lacks this property and the smoothness of the surface contributes to a less secure knot. Adjustment of tension with silk has a certain resistance easily gauged and controlled. Metallic or allied material may give way suddenly or partially crack as the tension is adjusted. For these reasons, fine ophthalmic silk has been more reliable in our hands than tantalum.

(d) Method of Introducing Suture Material. The method of introducing suture material is an obvious controlling factor which has been frequently overlooked. If multiple sutures are necessary, each new insertion or perforation is a traumatizing episode damaging nerve bundles and initiating bleeding. This constitutes a further reason for using as few sutures as possible. Trauma to the nerve ends is controlled by the mechanism of insertion as well as by the fineness of the suture material. To obtain advantage from fine suture material, the needle carrying it should be as small as the material itself. In addition to damage produced by needle and thread, simplicity of application is also a factor. Less control is possible when tying invisible sutures. This increased handling of nerve trunks produces more damage than when a slightly larger more visible material is used. It has seemed best to replace as many sutures as possible by a properly applied autogenous glue.

(e) Protection of Suture Line. Some form of insulation of the vital union tissue has long been felt to be of value. This was based on a desire to prevent infection extending into nerve ends and later causing strangling intraneural fibrosis. Advances in the control of infection no longer make this the main purpose of insulation, but some important considerations remain. Chief among these is some mechanism of immobilization at the suture line favouring the progress of axoplasm across the gap in oriented stream lines. A protec-

tive covering also minimizes extraneous stress or traction at the suture line and prevents creeping encroachment of fibrosis from adjacent structures. An autogenous plasma clot provides an immobilizing cuff which fulfills most of these desiderata. It serves as a further protection against irritation set up by silk or other foreign material necessary in the neurorrhaphy.

CONCLUSIONS

1. Plasma silk suture of peripheral nerves has proved practical in over 350 cases. The modified technic has made possible most advantages of an adhesive method.

2. Superior results have been obtained in all nerves with the most striking improvement occurring in the peroneal.

3. Improved primary treatment of wounds and early neurorrhaphy has allowed more extensive use than was originally felt possible. Some limitations in its use remain in awkward situations and in sutures under excessive tension.

4. The method outlined is simple and no complications have resulted. The moderate tension encountered in repairing nerves in gunshot wounds has been effectively controlled by few sutures supported by a clot.

5. Apart from its value in routine suture, special advantages have been found in applying the method to notch defects, branch lesions and small nerves.

6. The series may not be considered statistically significant but it demonstrates the possibilities of a trend towards sutureless re-union of nerve tissue.

7. The improved technical management afforded by the method enables precision accuracy. Following meticulous attention to the minatiae of each step in the suture process, more orderly regeneration may reasonably be expected.

The author is indebted to Dr. I. M. Tarlov for the generous experimental demonstration of his technic. Further appreciation is expressed to Dr. R. I. Harris, Dr. K. G. McKenzie, Dr. G. M. Dale and Dr. E. A. Linell, for their interest and guidance in the whole experimental and clinical program.

REFERENCES

[1] Bateman, J. E.: Evaluation of Plasma Suture of Nerves. Proc. National Research Council of Canada, 1945.

[2] Linell, E.: Pathology of an Experimental Investigation of Plasma Suture of Peripheral Nerves. Prov. National Research Council of Canada, 1945.

[3] Seddon, H. G., and P. B. Medawar: Fibrin Suture of Human Nerves. Lancet, **288**: 1942.

[4] Tarlov, I., and B. Benjamin: Plasma Clot and Silk Suture of Nerves. S. G. & O., **76**: 367–74, 1943.

[5] Tarlov, I. M., Denslaw, Swarzes, and D. Pineless: Plasma Clot Suture of Nerves. Archives of Surgery, **470**: 44–58, 1943.

[6] Tarlov, I. M.: Plasma Clot Suture of Nerves. Surgery, 257–69, 1944.

OPERATIVE EXPOSURE OF THE BLOOD VESSELS IN THE SUPERIOR ANTERIOR MEDIASTINUM *

HARRIS B. SHUMACKER, JR.,
NEW HAVEN, CONN.

FROM THE VASCULAR CENTER, MAYO GENERAL HOSPITAL, GALESBURG, ILLINOIS, AND THE DEPARTMENT OF SURGERY, YALE UNIVERSITY SCHOOL OF MEDICINE, NEW HAVEN, CONNECTICUT

ADEQUATE EXPOSURE is essential in any operation in the anterior superior mediastinum if damage to important blood vessels and nerves is to be avoided. The necessity for perfectly ample exposure is particularly important when operation is performed for an aneurysm or arteriovenous fistula of the mediastinal vessels. Almost every conceivable type of operative approach has been employed since Valentine Mott first ligated the innominate artery in 1818 in an effort to cure a traumatic subclavian aneurysm.[1]

In Greenough's excellent review of operations upon the innominate artery[2] the fact is brought out that safety can only be achieved by adequate resection of the overlying bony framework. In only 37 of the 91 cases of innominate ligation or attempted ligation was resection or osteotomy of the overlying bones performed, and it was apparent that inadequate exposure was the primary factor in a number of fatalities. An idea of the multiplicity of incisions is obtained from his study. In 11 cases a portion of a clavicle and of the manubrium was resected, while in 11 others only a part of the clavicle was excised, combined in one case with partial excision of the first costal cartilage. In the remainder, such procedures were carried out as sterno-clavicular disarticulation, mid-line splitting of the sternum, partial excision of the manubrium, clavicle and first rib, and resection of the manubrium with or without excision of the first two costal cartilages.

The type of operation in which exposure is accomplished by resection of bone is illustrated by the operation of Bardenheuer[3] in which the inner part of one clavicle and first rib are removed, the manubrium is sectioned transversely about an inch below its superior border, following which the opposite clavicle, first and second ribs are divided and the freed manubrium is excised. Various modifications of this operation have been used. Similar exposure is obtained by the osteoplastic operation of Kocher,[4] a procedure quite similar to that which had been advanced by Giordano and Auvray. The manubrium is reflected as a flap attached to the costal cartilages on one side after division or disarticulation of the clavicles, the first and second costal cartilages on one side, and transection of the sternum at the level of the second interspace. The exposure of Sauerbruch[5] has been widely used for certain mediastinal explorations and occasionally for vascular operations. It involves splitting of the sternum longitudinally down to the level of the third interspace and cutting it across into the third interspace.

Each case must be considered as an individual problem and an effort must

* Aided by a grant from the Office of Naval Research, the United States Navy.

be made to choose the ideal operative approach. The best type of incision for one is not necessarily the best for another case. Indeed the wide utility of different approaches is exemplified by the last three reported cases of innominate aneurysms and arteriovenous fistulas. Elkin[6] resected the inner half of the clavicle, the second costal cartilage and inner portion of rib, divided the manubrium and excised a portion of the right half of it. Trent[7] used a transpleural approach after resection of the second rib and cartilage and division of the third. Lindskog[8] excised the proximal portion of the second costal cartilage, rongeured partly across the sternum at this level, split the manubrium vertically with a Gigli saw introduced from above, and divided the clavicle.

Though aware of the value of various types of approach to the vessels in the superior anterior mediastinum, I have felt that it might be helpful to record my experience with one type of operative exposure which has proved exceedingly satisfactory in approaching aneurysms and fistulas involving a number of different mediastinal vessels. I shall include in this presentation brief case reports in order to demonstrate the general usefulness of this procedure. I had originally used, with slight modification, the operation of Sauerbruch in exposing an innominate aneurysm (Case 3). The sternum was split down to the level of the third interspace and across into both interspaces at this level. The exposure of the arch of the aorta, the proximal and mid portions of the innominate vessels and the main aneurysmal mass was excellent. However, in spite of wide retraction of the divided sternum, it was impossible to obtain adequate visualization of the subclavian and carotid arteries and the superior pole of the aneurysm which were hidden under the upper part of the manubrium and the sterno-clavicular joint. I have subsequently exposed several other large innominate aneurysms through the same incision. Again excellent visualization of the aneurysm and the origin of the innominate artery was obtained but the subclavian and, to a lesser extent, the carotid arteries were not satisfactorily visualized. In these cases the extreme dilatation of the innominate as it came off the aorta precluded, it was thought, ligation of the artery and treatment was confined to wiring and coagulation.

Because of this experience I have combined splitting of the sternum with resection of the inner third of the clavicle. Excision rather than transection of the clavicle has been performed because of considerations which have been presented elsewhere.[9, 10] This procedure has proved an excellent method for obtaining good exposure. An incision is made from the mid portion of the clavicle down over the sternoclavicular joint to the midline and is continued down over the sternum to the level of the third or fourth interspace. The platysma is divided. The inner third of the clavicle is resected subperiosteally by disarticulation of the sternal end and division of the other end with a Gigli saw. The sternal part of the sternocleidomastoid is divided as well as part of the clavicular origin of this muscle. The sternohyoid and sternothyroid muscles are similarly severed near their origin. Ordinarily it is necessary to divide these muscles only on the side from which the clavicle is removed. A finger is gently passed behind the manubrium from above, and it is generally

feasible to separate the mannbrium from the underlying structures in this manner. Occasionally a blunt dissector has been used cautiously in this dissection, but only with the constant guidance of the palpating finger in order to avoid trauma to the vessels beneath. The anterior periosteum of the sternum is incised in the midline and the sternum is split down to the level of the second or third interspace. The Schumacher sternal shears is ideal for this purpose; the Lebsche knife is also satisfactory though in my experience the bone tends to split in advance of the knife and not always in the midline. Once the sternum has been divided for some distance its edges can be retracted with bone hooks and one can palpate the underlying structures more satisfactorily and determine whether the sternal transection should be carried out at the

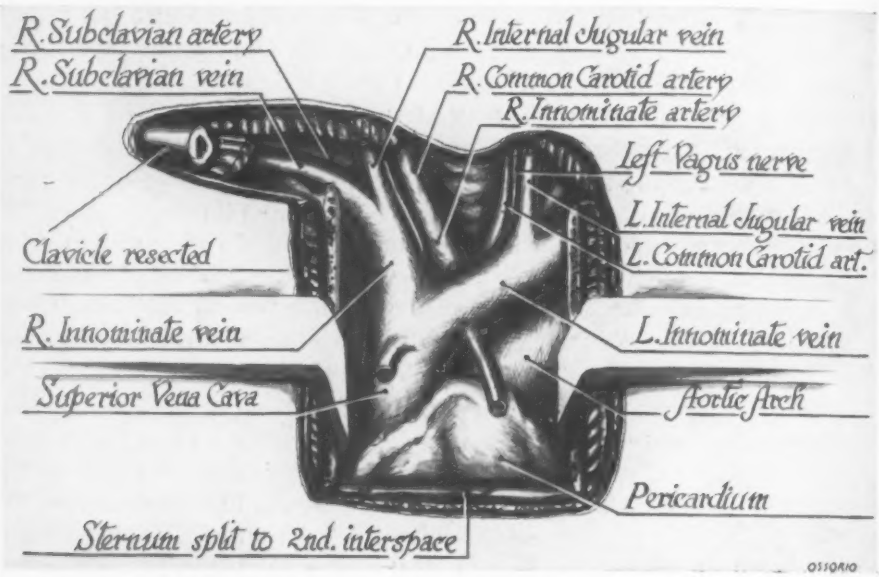


FIG. 1.—Illustration of the operative exposure. For the sake of simplicity the phrenic nerves, the thymus gland, the periosteum of the clavicle, the branches of the subclavian vessels and the divided right anterior scalene muscle are not shown.

second or third interspace in order to provide the desired exposure. One can also free more completely the sternum from the tissues beneath. The sternum is then divided across into both interspaces at the appropriate level after incising the periosteum across and freeing the outer borders. An ordinary rib shears can be used for this purpose. One now retracts the halves of the sternum widely, pushes the loose areolar tissue and thymus to one side or excises them, and brings into view the great vessels. A small rib spreader is substituted for the bone hooks.

The structures exposed are shown in a somewhat simplified semi-diagrammatic form in Figure 1. The innominate veins and the superior vena cava are in view. The arch of the aorta, the innominate, subclavian and carotid arteries are identified without difficulty. The phrenic nerves (which

are omitted in the drawing) and the vagi are usually readily seen. If it is required, the subclavian artery can be dissected out laterally following section of the anterior scalene muscle, care being exercised to avoid trauma to the phrenic. The first branches of the subclavian are ordinarily visible. In the drawing the sternal origin of the sternomastoid muscle on the side opposite that of the clavicular resection is shown divided; generally this is unnecessary. The drawing also shows the subclavian and carotid vessels on the side opposite that of the clavicular resection visible for a greater distance than is usually the case. In this illustration the sternum has been transected at the

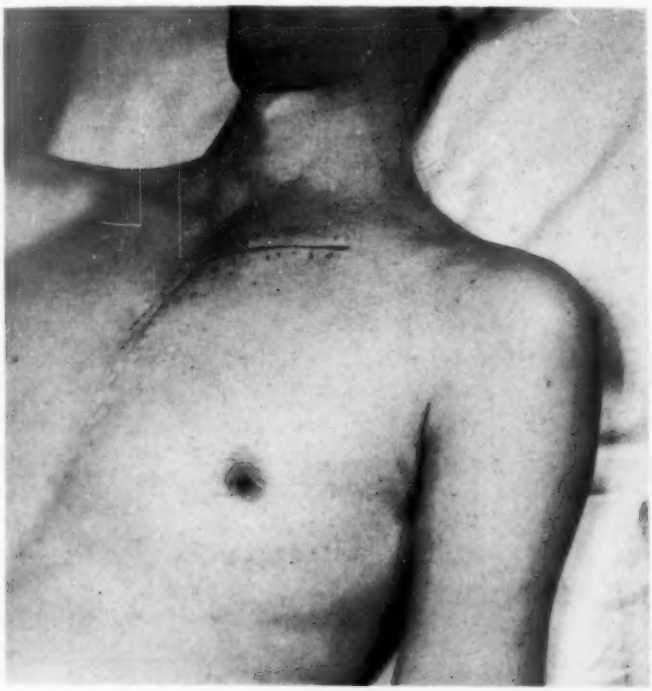


FIG. 2.—Wound two weeks after operation. (Case 6) The contour of the chest is relatively normal.

second interspace although sometimes division must be carried out an interspace lower in order to provide the same exposure. Indeed there is wide variation in the position of the mediastinal structures in relation to the overlying bony framework. Occasionally the arch of the aorta is as high as the superior border of the manubrium. Sometimes the pericardium is barely in view. At times the superior vena cava is seen to be of considerable length as in this drawing, while in other cases the innominate veins appear to join more caudally into a short vena cava.

Once the vascular surgery is accomplished, the wound is closed in layers. The sternum is re-approximated either with wire sutures placed through drill

holes or by silk sutures in the periosteum. The clavicular periosteal bed is carefully closed with interrupted silk sutures. In some cases the excised

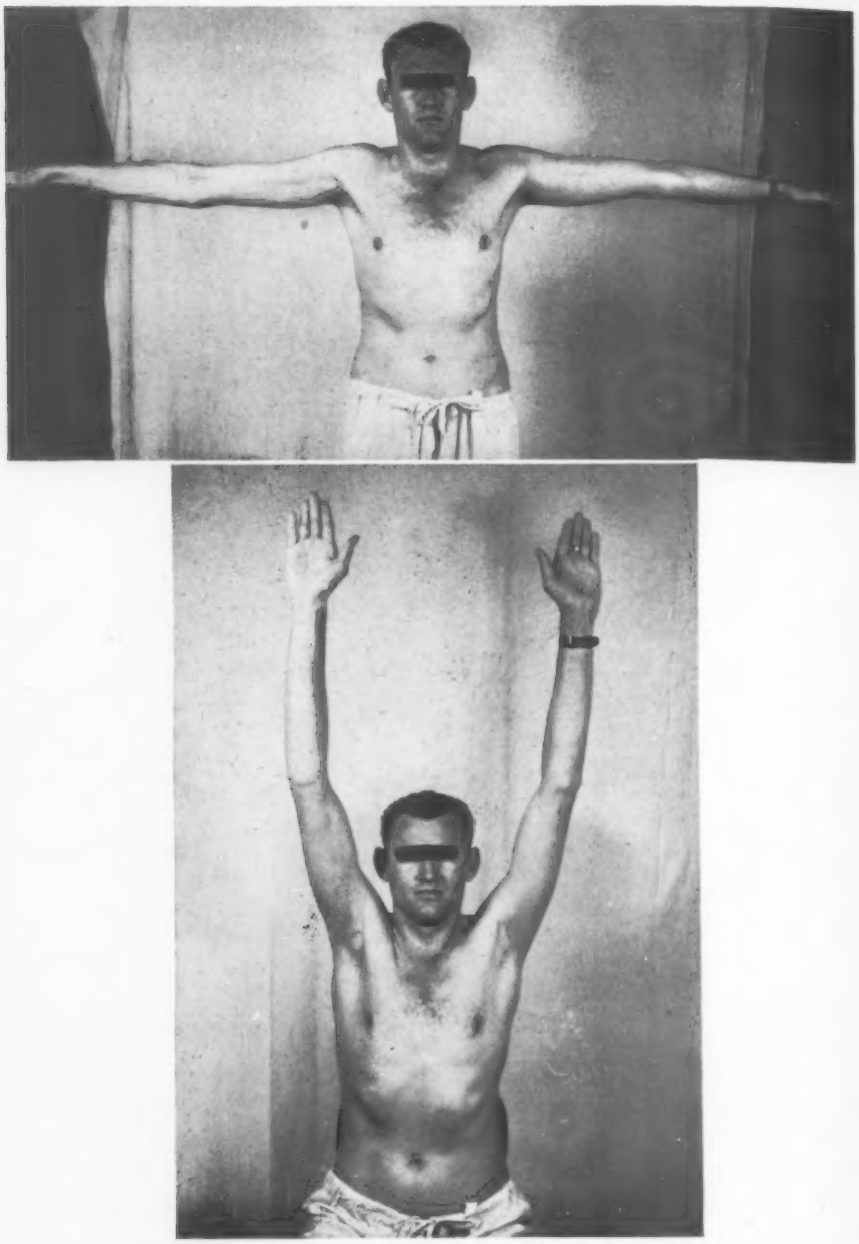


FIG. 3.—Photograph taken 2 months after operation showing normal range of shoulder motion. (Case 4)

portion of clavicle has been replaced as bone chips, a procedure which I feel is advisable since it hastens bony repair.[10] The muscles, fascia and skin are now

brought together with silk sutures. The skin incision is shown in Figure 2 and the normal range of shoulder motion after operation in Figure 3.

CASE REPORTS

Case 1.—The patient was a 35-year-old officer who had been wounded on August 19, 1944 by a small shell fragment which entered the right infraclavicular area near the sternum. In addition he received a shell fragment injury of the left thigh which resulted in a large avulsed wound. He was found to have signs of an arteriovenous fistula and was returned to the Zone of the Interior and admitted to the Mayo General Hospital.

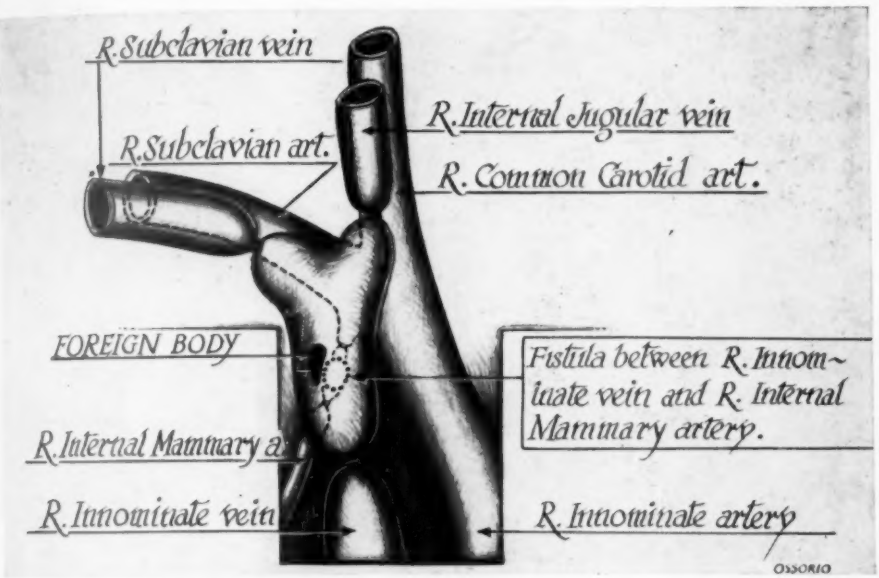


FIG. 4.—Condition found at operation in Case 1. Fistula between internal mammary artery and innominate vein. The point of ligation and division of the artery and vein are indicated.

He was aware of a buzzing, throbbing sensation in the right sternoclavicular area but had noted no other symptoms.

Examination revealed a continuous thrill and murmur most prominent just below the sternal end of the clavicle. The bruit was transmitted down towards the heart, out over the course of the subclavian and axillary vessels, and a little less well up into the neck. The thrill and bruit could not be obliterated by digital compression. Blood pressure was 108/58. There were no circulatory changes in the right upper extremity. Electrocardiogram was normal, roentgenograms negative except for the demonstration of a small fragment of the metal below the sternoclavicular joint.

On February 16, 1945 exploration was carried out. The condition found is shown in Figure 4. There was a fistula between the internal mammary artery and the overlying innominate vein. The shell fragment was imbedded in the posterior wall of the vein, which was indurated and scarred in this area. The fistula was excised with quadruple ligation of the vessels. Convalescence was uneventful and the patient has remained well.

Case 2.—The patient was a 24-year-old soldier who had been struck by shell frag-

ments on October 20, 1944, receiving penetrating wounds of the left malar region, left side of neck, and left supraclavicular fossa. The shell blast caused temporary loss of consciousness and loss of hearing in the left ear. He became hoarse shortly after injury. He was found to have a fracture of the maxilla which was reduced through a Caldwell-Luc approach, and a massive hemothorax which was treated by thoracentesis. Hearing began to improve but a distressing tinnitus persisted. He developed a throbbing, aching pain in the left arm and subsequently a persistent left ulnar hypesthesia. The hoarseness persisted. A decortication of the left lung was performed and he was admitted to the Mayo General Hospital on March 9, 1945, with a diagnosis of left carotid aneurysm. He complained of abnormal pulsation in the left side of the neck, throbbing pain in the left arm, numbness in left ulnar area, tinnitus in the left ear, watering of the left eye, and hoarseness.

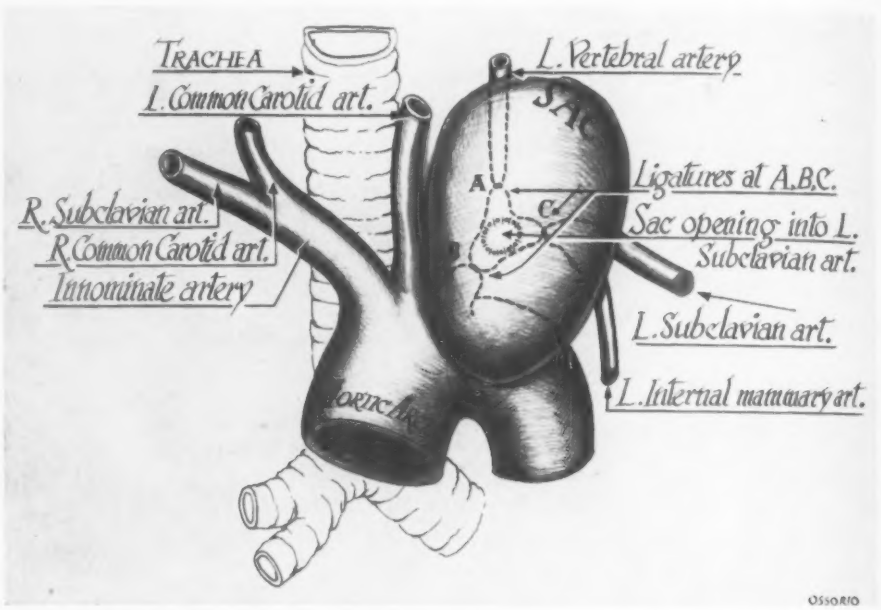


FIG. 5.—Condition found at operation in Case 2. Subclavian aneurysm. The points of ligation of the arteries are indicated

Examination showed a pulsating mass at the base of the left side of the neck without thrill but with a loud systolic bruit. Digital compression caused cessation of the bruit. Pressure over the inner part of the mass interrupted carotid pulsation and reduced the radial pulse; pressure over the outer part obliterated the brachial pulse. Blood pressure was 116/70. There was no circulatory disturbance of the left upper extremity. There was left ulnar hypesthesia, left recurrent laryngeal paralysis, and bilateral nerve-type deafness. Electrocardiograms were normal and roentgenograms not informative. There was evidence of good collateral circulation during the reactive hyperemia test with the left subclavian occluded. He stood prolonged left carotid compression without symptoms.

After a furlough and repeated digital occlusion of the carotid artery the patient was operated upon on May 4, with a diagnosis of traumatic aneurysm of the subclavian or common carotid artery. The condition found is illustrated in Figure 5. The left vagus nerve was stretched out over the large aneurysm, thinned but otherwise apparently uninjured. The carotid was uninvolved. The first portion of the subclavian was isolated

mesial to and underneath the sac and a tape placed about it. The subclavian was isolated distal to the sac. The vessel was dissected back towards the involved area beneath the aneurysm. When the artery was then occluded proximally and distally, the sac ceased to pulsate; it continued to fill rapidly, however, with bright arterial blood, as demonstrated by needle aspiration. It was now evident that the vertebral artery opened into the sac and it was feared that simple proximal and distal ligation of the subclavian would not effect a cure. Since the vertebral artery was inaccessible because of the overlying sac and adjacent scarring, the sac was carefully freed back to its mouth during temporary occlusion of the subclavian on either side, following which the sac was opened widely. Back bleeding was prevented by digital pressure while the subclavian was ligated just

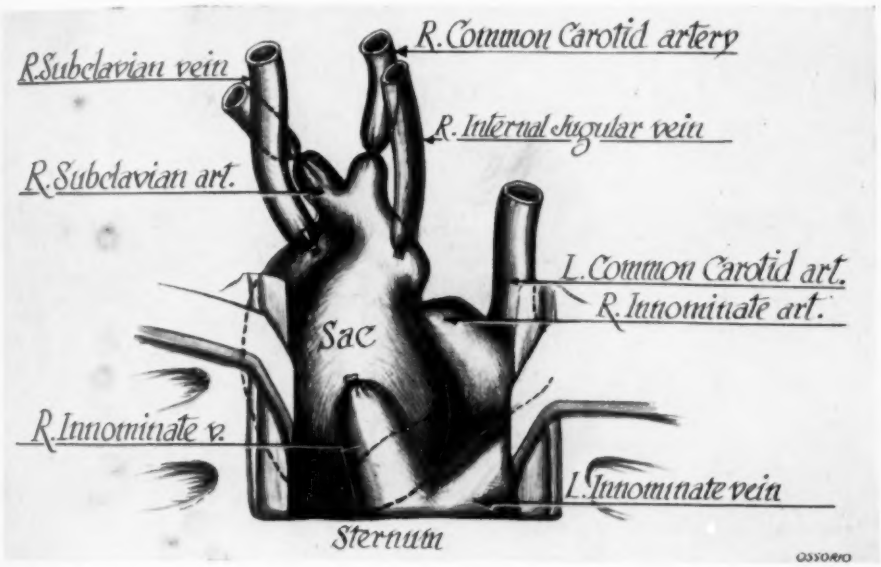


FIG. 6.—Condition found at operation in Case 3. Innominate aneurysm. The subclavian and carotid arteries were divided between ligatures and the sac was opened and emptied of its thrombus.

proximally and distally and the vertebral was dissected free and ligated. The sac was then resected subtotally. The overlying innominate, subclavian and internal jugular veins had been ligated and divided.

The patient withstood the procedure well but remained drowsy after operation. There was no paralysis of the extremities. About 8 hours later he complained of severe headache,—became stuporous, then comatose, and died about 22 hours after operation. Autopsy revealed extensive encephalomalacia of the left cerebellum. The circle of Willis was normal and there was no thrombosis of the left vertebral. No embolus was demonstrated.

Case 3.—This case is reported in detail elsewhere.[11] The patient was a 25-year-old soldier with a traumatic innominate aneurysm. Exploration was carried out 7 months after injury. Partial proximal ligation with a fascial band and cellophane was performed rather than complete ligation because marked ischemia of the hand was noted during temporary occlusion of the innominate artery. The reduction in oscillometry which followed was only temporary. A dorsal sympathectomy was carried out 3 weeks later and the mediastinum was explored again 3½ months after the original operation. After

ligation of the innominate, subclavian, and internal jugular veins and resection of the intervening segment, the innominate artery was ligated proximally, the subclavian and carotid arteries were divided between ligatures distally, and the sac was opened and evacuated of its thrombus (Fig. 6). Convalescence was uneventful and the patient has remained well, except for slight general weakness and a little fatigueability of the right hand.

Case 4.—The patient was a 27-year-old soldier who had received multiple injuries from shell fragments on January 28, 1945. He was wounded in the right thigh, the left buttock, the back, and the posterior aspect of his neck. After a time he was evacuated to the Zone of the Interior and admitted to the Mayo General Hospital with a diagnosis of right subclavian arteriovenous fistula. On admission he complained of a buzzing sen-

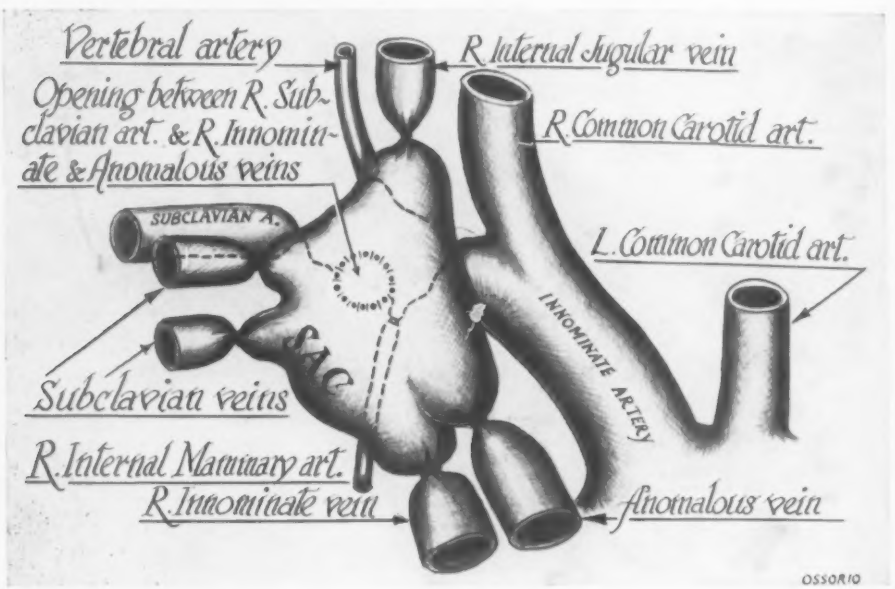


FIG. 7.—Condition found at operation in Case 4. Fistula between subclavian artery and innominate vein. The points of ligation or ligation and division of the various arteries and veins are shown.

sation in the right sternoclavicular area, numbness in the right ulnar distribution and hyperhidrosis of the right hand.

There was a continuous thrill centered over the sternoclavicular area and a continuous bruit so loud that it masked all respiratory sounds in the chest. The bruit and thrill could not be obliterated by digital compression. There was excessive sweating and slightly diminished temperature of the right hand, and slight reduction in oscillometry and blood pressure in the right upper extremity as compared with the left. Blood pressure was 142/76. Electrocardiogram was normal and roentgenogram not remarkable except for a metallic foreign body under the sternal end of the right clavicle. It was thought that the patient had an arteriovenous fistula involving the first portion of the subclavian vessels.

Exploration was carried out on June 13. The condition found is illustrated in Figure 7. The fistula existed between the origin of the subclavian artery and the innominate vein just where it received the internal jugular and two subclavian veins. A large anomalous

vein emptied proximally into the vena cava. It was possible to ligate the 5 veins involved, the subclavian artery just as it emerged from the innominate and just distal to the fistula, and the vertebral and internal mammary arteries which originated in proximity to the fistulous opening. The veins as well as the distal subclavian artery were divided between transfixing ligatures and a part of the venous sac was excised.

Convalescence was smooth and excellent circulation was maintained in the right upper extremity.

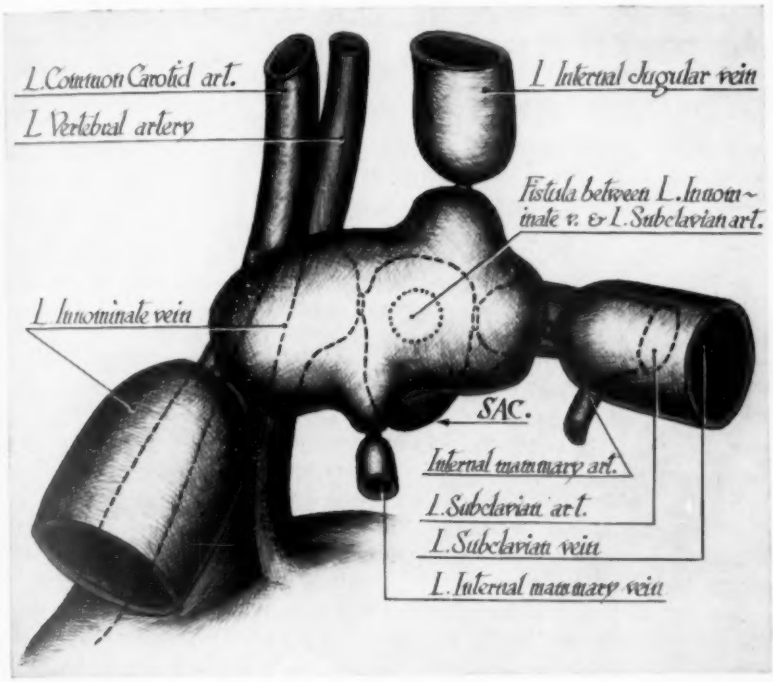


FIG. 8.—Condition found at operation in Case 6. Arteriovenous and saccular aneurysm of subclavian artery and innominate vein.

Case 5.—The patient was a 24-year-old soldier who on January 24, 1945, received shell fragment wounds of the abdomen, chest, hands, right forearm, arm and leg. The wounds were débrided, an exploratory laparotomy was performed, and numerous thoracenteses were done for bilateral hemothorax. He arrived at the Mayo General Hospital on May 21, 1945, with a diagnosis of arteriovenous fistula.

There was a continuous murmur audible over the upper anterior chest and maximal over the right border of the sternum at the level of the second interspace. No thrill was palpable. There were no circulatory disturbances of the extremities. Blood pressure was 124/60. Electrocardiogram was normal. Roentgenograms of the chest revealed a mass in the anterior superior mediastinum. The impression was that the patient had an arteriovenous and saccular aneurysm probably involving the innominate vessels.

He stood carotid compression without symptoms. It was impossible to close the fistula by compression and hence to test the collateral circulation to the right upper extremity. A dorsal sympathectomy was performed on July 5.

Mediastinal exploration was carried out on September 17. The upper portion of the

mediastinum was filled with a firm, pulsating aneurysmal mass about 7 by 6 cm. in size, associated with a continuous thrill. The carotid and subclavian arteries and the internal jugular veins could be identified at the upper pole of the mass on either side; they seemed to emerge from the aneurysm itself. The superior vena cava was free proximally but it disappeared into the mass about 2 cm. from its cardiac end. The superior border of the arch of the aorta was fused with and partly buried beneath the mass. The innominate vessels were not visible. It was felt that the arteriovenous and saccular aneurysm involved the innominate vessels, the origins of both carotid and subclavian arteries, as well as the terminal portions of the internal jugular veins, the superior vena cava, and possibly the arch of the aorta. Surgical extirpation appeared to be impossible. The wound was closed in the usual manner.

Convalescence was uncomplicated. He has remained in fairly good health and is able to do light work on a farm.

Case 6.—The patient was a 24-year-old soldier who had been injured in the left thigh, leg, arm and scapular area by shell fragments on December 10, 1944. Wounds were débrided and the left upper and lower extremities were placed in plaster casts. The femoral fracture progressed satisfactorily but osteomyelitis of the tibia and fibula at the site of the compound fracture was present upon admission to the Mayo General Hospital in May. An arteriovenous fistula in the left sterno-clavicular region had been recognized shortly after injury. There were no complaints referable to the fistula. There was weakness of the left upper extremity and an ulnar paralysis.

There was a continuous thrill and a loud continuous bruit over the left anterior chest wall and the left side of the neck, most prominent in the sterno-clavicular area. Neither bruit nor thrill could be obliterated by digital compression. The left fingers were cooler than the right and pulses and oscillometric readings were much reduced in the left upper extremity as compared with the right. After a period of treatment on the orthopedic service, and left ulnar neurorrhaphy, the patient was returned to the vascular service on August 22. Because of the evidence of reduced circulation in the left upper extremity and the impossibility of testing the collateral circulation, a dorsal sympathectomy was performed on August 30.

On September 18 mediastinal exploration was undertaken with a preoperative diagnosis of arteriovenous fistula of the proximal portion of the subclavian vessels. The condition found is illustrated in Figure 8. There was a fistula between the subclavian artery just distal to the vertebral branch and the innominate vein near its distal end. The vessels were isolated proximally and distally and the fistula was dissected free and transfixed. It was then discovered that there was in addition a saccular aneurysm arising from the posterior surface of the artery opposite the fistulous opening. The artery was badly damaged over a considerable distance. Consequently the artery was ligated on either side of the aneurysm, the innominate, internal jugular, subclavian and internal mammary veins were ligated and divided, and the specimen was excised.

Convalescence was uneventful and the patient was returned to the orthopedic service for further treatment for the osteomyelitis in the leg.

SUMMARY AND CONCLUSIONS

1. In any contemplated excision of an aneurysm or arteriovenous fistula it is essential to isolate the vessels proximal and distal to the lesion before directly attacking the lesion itself. This necessitates ample exposure.

2. Vascular operations upon the vessels in the anterior superior mediastinum require resection or division and retraction of the overlying bony framework.

3. An operative procedure which has given excellent exposure without

injury to the underlying structures and which yields a good cosmetic and functional result is presented and is illustrated by several case reports.

4. Although each case must be individualized and the proper operative approach selected, it is believed that the procedure outlined will be found suitable for many explorations in the anterior superior mediastinum.

REFERENCES

1 Mott, V.: Reflections on Securing in a Ligature the Arteria Innominata. Med. & Surg. Register, **1**: 9, 1818.

2 Greenough, J.: Operations on Innominate Artery. Report of a Successful Ligation. Arch. Surg., **19**: 1484–1544, 1929.

3 Bardenheuer: Die Resection des Manubrium Sterni. Deutsche. Med. Wshnschr., **11**: 688–690, 1885.

4 Kocher, T.: Textbook of Operative Surgery. Vol. II. New York, The Macmillan Co., 1911.

5 Sauerbruch, F., and E. D. Schumacher: Tecknik der Thoraxchirurgie. Berlin, J. Springer. 1911.

6 Elkin, D. C.: Arteriovenous Aneurysm. The Approach to the Innominate Vessels. J. A. M. A., **129**: 26–27, 1946.

7 Trent, E. R.: Ligation of the Innominate Artery for Innominate Aneurysm using Rubber Bands: Report of a Case. Surg., Gynec. & Obst., **82**: 463–470, 1946.

8 Lindskog, G. E.: The Surgery of the Innominate Artery. New England J. Med., **235**: 71–76, 1946.

9 Shumacker, H. B., Jr.: Incisions in Surgery of Aneurysms with Special Reference to Exploration in the Anticubital and Popliteal Fossae. Ann. Surg., **124**: 586–598, 1946.

10 ————: Resection of the Clavicle with Particular Reference to the Use of Bone Chips in the Periosteal Bed. Surg., Gynec. & Obst., **84**: 245–248, 1947.

11 ————: Surgical Cure of Innominate Aneurysm. Report of a Case, with Comments on the Applicability of Surgical Measures. Surgery, **22**: 729–739, 1947.

BRONCHIOGENIC CYSTS OF THE MEDIASTINUM

HERBERT C. MAIER, M.D.
NEW YORK, N. Y.

FROM THE THORACIC SURGICAL SERVICE, MEMORIAL HOSPITAL, NEW YORK

A VARIETY OF CYSTS occur in the mediastinum and the vast majority are congenital in origin.[6] Some arise as a result of developmental aberration of the primitive foregut. The term bronchiogenic is usually applied to cysts arising from the respiratory system, whereas those derived from the digestive tract are termed esophageal or gastric. There is no sharp line of distinction between the bronchiogenic cysts and those arising from the digestive tube. This paper, nevertheless, will be concerned chiefly with the bronchiogenic group. The subject of intrathoracic cysts arising from the digestive tract has been recently discussed by Schwartz and Williams[35] and by Ladd and Scott.[20] Bronchiogenic cysts of the mediastinum are closely related to aberrant pulmonary tissue[11] and congenital intrapulmonary bronchial cysts.[15] As some of the clinical features as well as the therapeutic problems of bronchiogenic cysts of the mediastinum may differ from the intrapulmonary cysts, it seems advisable to consider the former as a separate group. The intrapulmonary bronchiogenic cysts will not be considered in this paper, as the subject has been previously discussed elsewhere[22, 23] and reviewed by Pugh.[29]

Until recently bronchiogenic cysts of the mediastinum have been considered to be rare, but with more frequent roentgenologic examinations of the thorax and the widening scope of thoracic surgery, many more cases of this type are being observed. Although some authors have believed the bronchiogenic mediastinal cyst to be usually asymptomatic, an analysis of all the case reports, both in the pathologic and surgical literature, suggests that a considerable number eventually cause symptoms of varying degree. Occasionally a bronchial cyst of the mediastinum causes death in early life by compression of the trachea or main bronchi.

Bronchiogenic cysts of the mediastinum which do not cause symptoms in early life may be found by chance on roentgenographic examination in adult life, or may be an incidental finding at autopsy. In some instances, however, due to the gradual increase in the size of the cyst with resultant pressure on adjacent structures, symptoms of varying degree may lead to clinical investigation and diagnosis. During a two-year period (1943–1944) five patients with a bronchiogenic cyst of the mediastinum were operated upon by the author on the Thoracic Surgical Service of the Memorial Hospital and three additional cases have been operated upon elsewhere.* The eight cases illustrate various features of bronchiogenic mediastinal cysts and some problems in the surgical therapy of this lesion. In addition to reporting these eight cases, the literature of mediastinal cysts is reviewed and the collected cases have been analyzed.

* Two cases at Lenox Hill Hospital and one case at Kings County Hospital.

ORIGIN OF MEDIASTINAL BRONCHIOGENIC CYSTS

A brief discussion of the embryology of the primitive respiratory tract may clarify the problems associated with the development of bronchiogenic cysts.[3] The respiratory tract has a common origin with the esophagus from the primitive foregut. As a result of the lateral invasion of two septa, the foregut is divided into a ventral and a dorsal component. These two masses of cells eventually are separated from one another and the dorsal component forms the esophagus, while the ventral component forms the trachea and major bronchi. The close embryological association of the respiratory tract

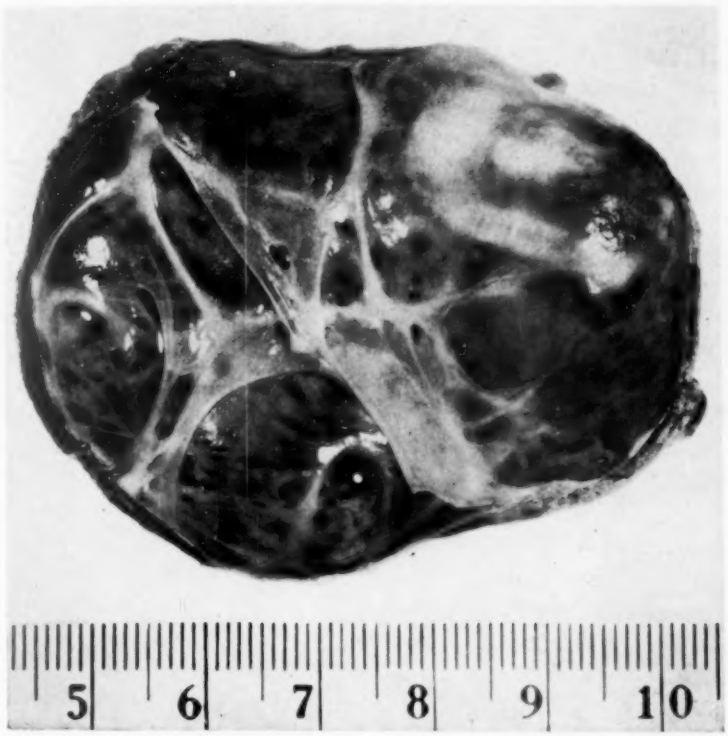


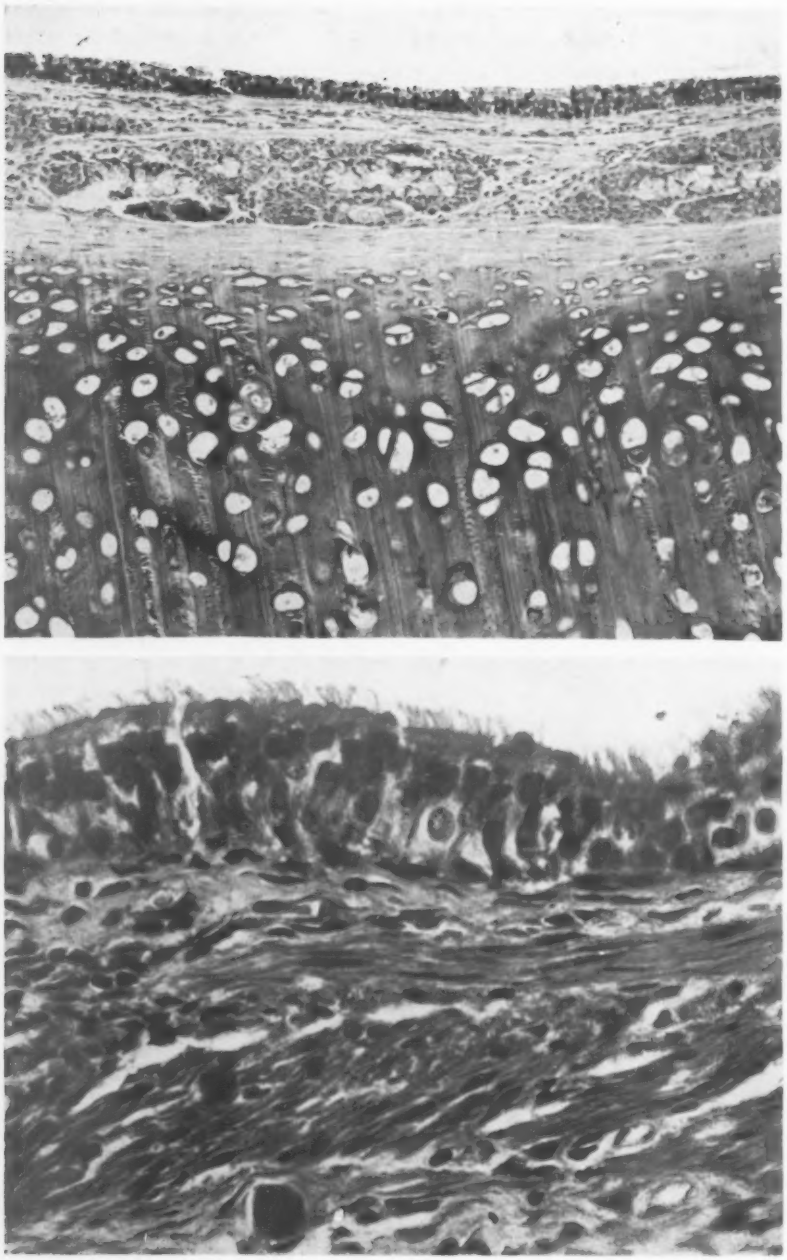
FIG. 1.—Bronchiogenic cyst. Appearance of inner aspect after evacuation of fluid contents. Note the thin wall and the numerous trabeculations.

with the primitive foregut indicates the possibility of a close association of developmental anomalies of these two structures.

Bronchiogenic cysts result from an abnormal budding or branching of the tracheobronchial tree.[34] If the continuity with the bronchial tree is maintained, the cyst is usually intrapulmonary or in intimate association with the lungs. If the mass of cells becomes separated from the tracheobronchial tree, there may be no continuity with the bronchial lumen. Such cysts gradually increase in size because of the distension produced by the secretion within the cavity.

An analysis of the literature reveals a difference of opinion concerning the

A



B

FIG. 2.—A. Photomicrograph of wall of bronchiogenic cyst. Appearance similar to wall of a bronchus. Ciliated columnar epithelium, mucous glands, cartilage, and muscle fibers are present.

B. Higher magnification of cyst wall clearly demonstrates the ciliated columnar epithelium.

stage in embryonic development at which aberrations occur which result in bronchiogenic mediastinal cysts. Although the wall of a typical bronchiogenic cyst is composed of structures similar to that of the bronchial wall, in an occasional case components of the digestive tract are also found in the same cyst wall.[37] The latter finding has been advanced as an argument in favor of the contention that the abnormal development begins at an early stage of the embryo before the separation of the respiratory tract from the primitive foregut. Whereas the majority of bronchiogenic cysts are found in relationship to the trachea or main bronchi, in an occasional instance the cyst may be in close association with the esophagus or even the vertebral bodies. These findings lend support to the contention that in at least some instances the misplacement of cells occurred at a relatively early stage of development.

PATHOLOGY

Bronchiogenic cysts usually appear as a single spherical cystic mass, but the cyst may be lobulated, or occasionally there is a chain of cysts. On section the cysts are often single, but may be multiloculated or contain several noncommunicating cavities of varying size. The inner aspect is frequently trabeculated (Fig. 1). The cyst wall is usually relatively thin, although there may be thicker portions. The cyst is lined by ciliated columnar epithelium and the wall contains mucous glands, cartilage, elastic tissue and smooth muscle (Fig. 2A and B). In some instances the cartilage or mucous glands may be absent. The uninfected cyst usually contains a thick, white mucoid material. Even in the absence of infection the cyst contents may be dirty brown[40] and sometimes closely resembles thick, purulent material. The cyst only rarely has any actual communication with the lumen of the tracheobronchial tree. Secondary infection may produce an inflammatory process which results in destruction of the lining epithelium and may render definite recognition of the bronchiogenic origin uncertain, especially if cartilage is not present in the cyst wall. The cyst may have a definite attachment to a portion of the tracheobronchial tree, but in some instances, although it is adjacent to a bronchus, there may be only loose areolar tissue between the two structures. Occasionally, a bronchiogenic cyst has no apparent connection with the tracheobronchial tree. If the bronchiogenic cysts of the mediastinum are segregated according to their location, certain embryologic, pathologic, and clinical features characteristic of each group may be noted. Bronchiogenic cysts may be arbitrarily divided into the following groups:

1. Paratracheal
2. Carinal
3. Hilar
4. Paraesophageal
5. Miscellaneous

1. *Paratracheal Group.* Several instances of a bronchiogenic cyst attached to the tracheal wall have been reported. A characteristic example of this type

of cyst is illustrated by Case 1. The cyst is usually attached to the right lateral tracheal wall a short distance above the bifurcation of the trachea. The site of attachment of the cyst is similar to the point of origin of the first lateral bronchus of some mammals. Moreover, diverticuli of the trachea have been reported in this same location.[7] The evidence suggests that bronchial cysts of the trachea occur as a persistence of a structural characteristic of lower forms. The embryology of the tracheobronchial tree in the pig has been thoroughly studied by Flint.[10] He has demonstrated that the lateral bronchial branch from the trachea ordinarily occurs only on the right side. The instances of bronchiogenic cysts in which the cyst had an intimate association with the tracheal wall have almost all been on the right side. An analysis of these data leads to the conclusion that this type of bronchiogenic cyst is explained on the basis of the rudimentary development of a first lateral bronchial branch from the trachea. Blackader and Evans[4] reported a paratracheal bronchiogenic cyst which compressed, but was not attached to, the left side of the trachea, and which caused the death of a nine-months-old boy.

2. *Carinal Group.* A number of cases in which the bronchiogenic cyst was located near or just beneath the bifurcation of the trachea have been reported. In some instances there was a definite strand-like attachment extending to the carina.[18] In other cases the attachment was to one of the larger bronchi, whereas in still others the attachment to the bronchial tree was not definitely demonstrable. The frequency with which small bronchial cysts occur in the subcarinal area suggests the possibility of a relationship with other types of congenital anomalies. The bronchiogenic cysts often have a definite attachment to the carina. In some instances there is also an attachment to the anterior esophageal wall, or the cyst may be in the esophageal wall in this region. The location of these cysts corresponds closely with the most common site of congenital tracheo-esophageal fistula. The frequency of embryologic aberrations in this region is striking. The subcarinal bronchiogenic cysts may well represent a pinching off of a group of cells in the communicating channel between the primitive foregut and tracheobronchial tree. The bronchiogenic cyst arising in the carinal area may cause pressure on either the right or left main bronchus, or both.[25] If the cyst is large, it may also press on the lower trachea. The cyst may be relatively small and still cause considerable compression of the air passage, due to its rather confined location. Most of the reports of this type of cyst are to be found in the pathologic literature because the diagnosis was unsuspected before death.[12] The majority of the patients died within the first year of life, due to pulmonary complications secondary to compression of the tracheobronchial tree. Although no successful surgical removal of a bronchiogenic cyst in this location in an infant has been reported to date, the lesion should be amenable to surgical therapy if the correct diagnosis were made. In a discussion of the clinical features of this group of bronchiogenic cysts, the clinical picture which should suggest the possibility of the existence of the lesion will be outlined. Whereas some cysts in the same location, chiefly due to their small size, have been chance findings

at autopsy in late adult life, a large percentage of the cases that have been published have caused serious, if not fatal results from secondary pulmonary complications.

3. *Hilar Group.* The majority of bronchiogenic cysts which are diagnosed in the later part of childhood or in adult life are located in the hilar area and have an attachment to one of the main or lobar bronchi. In some instances the attachment to the bronchus is well defined, whereas in other cases only connective tissue strands bind the cyst wall to the bronchus. As the cyst increases in size, it projects into the pleural cavity.[24]

Because of its close proximity to the main bronchus, it may cause compression of the main stem or one of the lobar bronchi. The secondary pulmonary changes which may occur will be described under the clinical findings. A considerable portion of a large cyst may be in close proximity to the pericardium and the cyst may project between the various hilar structures of the lung. In one of my patients there was an anomalous distribution of the hilar structures of the right lower lobe and the cyst was intimately associated with these structures and displaced them to a considerable degree. In this case there was an extension of the cyst across the mediastinum almost into the opposite hemithorax.

The bronchial cysts occurring in the hilar area are embryologically closely related to aberrant pulmonary tissue and anomalous accessory lobes of the lung. A review of the subject of aberrant intrathoracic pulmonary tissue has been presented by Friedlander and Gebauer.[11]

Secondary infection of the bronchiogenic cyst may result in considerable destruction of the cyst wall, rendering identification difficult. Moller[27] has reported a case of bronchiogenic cyst which ruptured into the tracheobronchial tree with the development of secondary infection. The cyst was in an unusual location, in that it lay in the anterior mediastinum. At operation the communication between the cavity of the cyst and the bronchial tree could not be demonstrated.

4. *Paraesophageal Group.* Some bronchiogenic cysts may be in close relationship with the esophagus and have little or no apparent connection with the tracheobronchial tree. In some instances the bronchiogenic cyst is merely in close proximity or loosely attached to the wall of the esophagus. In other instances, however, the cyst is entirely within the walls of the esophagus and bulges into the esophageal lumen, only the mucous membrane of the esophagus covering the inner aspect of the cyst wall. If an intramural esophageal cyst is lined by ciliated columnar epithelium but contains no cartilage within its wall, the bronchiogenic origin may be seriously questioned.[26, 19] Cysts of this type located in the lower esophagus near the cardia have been reported by v.Wyss,[39] Zahn,[42] Rau,[30] Tresp,[38] and Westenryk.[41] Robbins[32] reported a bronchiogenic cyst within the wall of the esophagus which contained cartilage, and was lined by ciliated columnar epithelium. A number of cases have also been reported in which a cyst lined by ciliated columnar epithelium was found attached to the outer portion of the esophagus. In several of these same cases

a well-defined double muscle layer was present in the wall of the cyst.[9] In one of my cases (Case 2) the cyst extended for a considerable distance parallel with and to the right of the esophagus. There were three separate cyst cavities extending from the level of the thoracic inlet down beneath the arch of the vena azygos to the level of the pulmonary hilum. The paraesophageal location of the cyst in this case, as well as the thick double layer of muscle in the wall, suggested a re-duplication of the alimentary tract. Moreover, no cartilage was found in the cyst wall. Although the lining of the cyst was typical ciliated columnar epithelium suggesting a bronchial origin, the presence of ciliated columnar epithelium lining the esophagus at one phase of embryological development makes the matter inconclusive.

5. *Miscellaneous Group.* Occasionally a bronchiogenic cyst is found in a very unusual location. Two cases have been reported in which the cyst was found within the pericardium.[17, 28] Rusby and Sellors[33] reported a case of bronchiogenic cyst associated with a congenital deficiency of the pericardium. The patient had symptoms which were attributed to the bronchial cyst. This was removed at operation which was followed by an uneventful recovery. A defect in the pericardium was also present in one of my cases (Case 7). The bronchiogenic cyst was located anteriorly in close proximity to the site of congenital absence of a portion of the upper part of the pericardium. Guillery[13] found a cyst lying on the anterior aspect of the thoracic spine and extending into the vertebral bodies, together with a smaller cyst of similar type behind the vertebral bodies. In this case no cilia were demonstrated on the cylindrical epithelium which lined the cyst. The muscle layers were more suggestive of the wall of the digestive tract than that of the respiratory system. The infant had died at the age of three months. The portion of the cyst which projected forward from the thoracic vertebrae had caused compression of the bronchi, especially on the right side.

Seybold and Claggett[36] reported a presternal cyst lined by ciliated, pseudocolumnar epithelium, which was located in the subcutaneous tissues at the sternal angle. The walls were thin and contained mucous glands, cartilage, vessels and nerves. This cyst bore a striking similarity to a bronchiogenic cyst of the mediastinum, and it was considered that the cyst had migrated into a presternal position. The underlying sternum showed no abnormality.

SYMPTOMATOLOGY

In the absence of infection, the symptomatology of bronchiogenic cysts depends chiefly on the size and location of the mass. In some instances there are no symptoms referable to the lesion, and the presence of the cyst is only demonstrated by a routine roentgenogram of the chest, or is a chance finding at autopsy. In general, the symptoms presented are those produced by compression of the tracheobronchial tree. In some of the patients in whom a bronchiogenic cyst was removed because of the finding of an undiagnosed mass

on a roentgenogram, the symptoms were rather vague and not characteristic, so that without a follow-up study it is difficult to ascertain whether or not the symptoms were caused by the cyst. When secondary infection supervenes, the symptoms resemble those of intrathoracic suppuration, particularly those of mediastinal or pulmonary abscess. Occasionally there is prominence of the chest wall in the region of the cyst. The symptomatology in special groups will now be discussed.

1. *Paratracheal Group.* In the few cases of this type that have been reported, no characteristic symptoms have been present. One case had previously had an empyema on the same side, but at the time of operation ten years later no infection was present. Due to its location, the cyst may cause considerable narrowing of the trachea, as is illustrated in Case 1, but the compression is usually insufficient to cause serious obstruction.

2. *Carinal Group.* When the bronchiogenic cyst is located just beneath the bifurcation of the trachea and causes symptoms in early life, the history may indicate some respiratory difficulty at or shortly after birth. In other cases nothing abnormal is noted until a respiratory infection develops. The natural tendency is to ascribe the symptoms to the pulmonary infection. Sometimes without such an evident precipitating factor, difficulty in breathing may be noted. Clinical and roentgen examination may indicate either obstructive emphysema or atelectasis, depending upon the degree of bronchial obstruction. Wheezing may be present; a croupy cough is sometimes noted. Varying degrees of dyspnea and cyanosis may develop. Progressive respiratory difficulty ensues and frequently results in a fatal outcome.[2] Analysis of the literature suggests that in those cases in which the subcarinal type of cyst causes symptoms in early life, the mortality rate is extremely high.

In some infants noisy breathing or attacks of cyanosis, especially during crying, may have been noted since birth. At times expiratory stridor is present. If the wheezing has been present for a considerable period of time, an asthmatic condition may erroneously have been considered the explanation of the symptoms. Adams and Thornton[1] have reported a bronchiogenic cyst in this location which first caused symptoms in adult life when secondary infection supervened. Robbins[32] reported a bronchiogenic cyst in an 18-year-old male which was located just beneath the carina. This cyst measured 8 x 6 cm. and caused an irritating, non-productive cough apparently due to pressure on the main bronchus. Surgical removal was successfully carried out. Similar cysts have occasionally been chance findings at autopsy in adults.[31]

3. *Hilar Group.* Bronchiogenic cysts arising in the area of the pulmonary hilum usually project a varying amount into the pleural space on that side. These lesions are often asymptomatic, but when symptoms are present, these are usually due to compression of a portion of the bronchial tree. Dull chest pain, dry cough, wheezing and frequent respiratory infections may occur. In occasional cases, such as in Case 4, moderate bronchial compression over a considerable length of time may cause chronic pulmonary infection which causes a productive cough. This same patient had slight discomfort on swal-

lowing, due to displacement of the esophagus by the cyst. In one of my patients there were no complaints prior to the accidental finding of the bronchiogenic cyst on a routine roentgenogram of the chest. Following removal of the cyst, however, the patient gained considerable weight and felt far better than he had for many years. Undoubtedly in this case the cyst was causing definite although unrecognized disturbance, probably due to the secondary changes in the lung which, however, had failed to cause cough or expectoration.

4. *Paraesophageal Group.* When the bronchiogenic cyst occurs in close relationship with or within the wall of the esophagus, there may be symptoms referable to that organ. Some dysphagia may be noted. The symptoms are similar to those encountered in benign tumors within the wall of the esophagus. Occasionally the lumen of the esophagus above the lesion is dilated. If the cyst is chiefly outside the wall of the esophagus, the patient may be asymptomatic.

5. *Miscellaneous Group.* In the cases of bronchiogenic cyst of the mediastinum occurring in miscellaneous locations, the only characteristic symptom complex is that which occurred in the two cases of intrapericardial cysts. These patients died suddenly, apparently owing to pressure on a portion of the heart or great vessels.

DIAGNOSIS

Until the last few years a diagnosis of bronchiogenic cyst has rarely been made before operation or autopsy. In some instances the clinical and roentgenologic picture presents nothing sufficiently characteristic to differentiate the lesion from other types of mediastinal tumor. There are, however, certain clinical and roentgenologic features which may lead to either a probable, or even in some instances, a definite diagnosis prior to operation.

1. *Paratracheal Group.* As the clinical findings in this group are not characteristic, the roentgen examination would seem to be the sole evidence upon which a diagnosis might be made prior to operation. Bronchiogenic cysts would have to be differentiated from other tumor masses in the upper mediastinum lying in close proximity to the trachea. A substernal thyroid, a thymic tumor, an intrathoracic hygroma, a serous cyst and an aneurysm are the chief lesions to be considered in a differential diagnosis. A substernal thyroid often surrounds the trachea to a greater extent than a tracheal cyst, which is usually located entirely to the right of the trachea. The borders of the mass as seen radiographically are generally more sharply defined in a cyst, as contrasted to a soft tissue mass such as a thyroid adenoma. Both lesions, however, may show calcification.

A thymic tumor may have a less well defined border on the roentgenogram, usually projects bilaterally from the mediastinum and is located more anteriorly. The upper limit of a paratracheal cyst will usually be above the level of the clavicle on the roentgenogram, whereas a thymic tumor begins below the clavicle and extends further downwards.

Differentiation of a bronchiogenic cyst attached to the trachea from an intrathoracic hygroma or serous cyst may offer real difficulty. If aspiration is performed, thick white or yellow mucoid material may be obtained from the bronchiogenic cyst in contrast to the clear watery fluid in the hygroma or serous cyst.

An aneurysm, particularly of the innominate artery, might give a somewhat similar roentgen appearance. Angiocardiography may definitely establish whether the mass is of vascular origin. An aneurysmal sac filled with a laminated clot, however, will not be delineated by the opacified blood, and therefore this diagnostic procedure does not always give positive information, particularly if the degree of opacification is somewhat unsatisfactory.

2. *Carinal Group.* The diagnosis of the subcarinal bronchiogenic cysts, especially when producing symptoms in infants, has apparently been very difficult in the past. As mentioned in a discussion of the symptomatology, the picture is essentially that of a bronchial, or occasionally lower tracheal, obstruction. It is usually erroneously assumed that the obstruction is produced by an inflammatory process within the air passages, whereas actually the pulmonary infection is secondary to bronchial compression by the cyst from without. A roentgenogram of the chest may give no indication of the presence of a mediastinal tumor because the cyst is relatively small and is completely hidden in the mediastinal densities. If the possibility of a cyst were borne in mind, an oblique film might be helpful in delineating the mediastinal mass, provided the superimposed densities from the pulmonary infection do not cause obscuration.

A clue to the true nature of the lesion may be obtained by bronchoscopic observation in certain cases. If bronchoscopy shows no endobronchial lesion but external compression of the lower trachea, or one or both main bronchi, a mediastinal tumor of some type must be ruled out. Other lesions, however, may also cause extrinsic pressure on the lower trachea. Congenital anomalies of the large vessels may be associated with obstruction to trachea or esophagus, or both. Operative intervention might be advisable in either situation.

3. *Hilar Group.* Apparently the most frequent preoperative diagnosis in patients with bronchiogenic cyst reported in the literature has been mediastinal dermoid. A point which should be of considerable aid in differentiating these two lesions is that the dermoid tumors arise in the anterior mediastinum, whereas the bronchial cysts are more frequently located in the posterior mediastinum. A few dermoid cysts have been reported as arising in the posterior mediastinum, but on reviewing these cases, most of them are found to be instances of bronchiogenic cysts rather than mediastinal dermoid.[14] In some instances bronchiogenic cysts have been confused with neurogenic tumors. On the lateral roentgenogram neurogenic tumors are seen to be located in the costovertebral portion of the thorax, whereas the bronchiogenic cysts are in the posterior mediastinum anterior to the vertebral bodies.

Robbins[32] has recently reported the roentgenologic findings in several cases of bronchiogenic cysts. He points out that bronchiogenic cysts may move

with respiration and also may change shape with respiration, indicating their cystic nature and relationship with the tracheobronchial tree. This finding may be of aid in differentiating bronchiogenic cysts from certain other mediastinal tumors. Dermoid cysts have a rather stiff wall and usually do not change contour with respiration. Included in Robbins' report are several cysts in which the histopathologic findings do not definitely demonstrate the origin of the cyst. The author included these cases because he thought they may be of value in linking the other cases of so-called simple or unclassified cysts of the mediastinum into one group. Whether these other cases are actually of similar origin to true bronchiogenic cysts cannot be stated at this time. In two of the cases the cyst was located in the anterior mediastinum in close relationship to the pericardium and diaphragm. In neither of these cases was the cyst wall entirely typical of a bronchiogenic cyst, and the radiologic and histologic findings suggested the possible diagnosis of pleural cyst of the type discussed by Lambert.[21] Also included in the report of Robbins are cases of bronchiogenic cysts within the substance of the lung and an intrapulmonary cyst-like lesion secondary to chronic abscess, which are not to be confused with the cases considered in the present paper.

Rarely a mediastinal bronchiogenic cyst perforates into the lung, with resultant bronchial communication. Then the roentgenologic and clinical picture simulates either a mediastinal or pulmonary abscess. As bronchiogenic cysts are closely related to aberrant pulmonary tissue, a cyst-like cavity lined with respiratory epithelium may be formed as a budding from the bronchial tree. Harrington[15] has reported a case in which the azygos lobe consisted of a sac similar to the wall of a bronchiogenic cyst. There was a small bronchial communication with the upper lobe. The connection with the tracheobronchial tree permitted the partial evacuation of the cyst contents and resulted in the presence of a fluid level within the cyst cavity at one roentgenographic examination. A few other instances of bronchiogenic cyst of the mediastinum with a fluid level have been reported, but this finding usually indicates that the cyst is intrapulmonary.

4. *Paraesophageal Group.* A bronchiogenic cyst may be in close association with the esophagus, or even the cardiac portion of the stomach. The lesion must be differentiated from solid tumors of the esophageal wall.

A considerable number of cases are on record in which a cyst lined by ciliated epithelium has been found within or in close association with the wall of the esophagus. In the majority of instances no cartilage has been found in the cyst wall, and therefore it is questionable whether they should be classified with the bronchiogenic group. Those cases without cartilage might be considered as having resulted from the pinching off of the cells from the esophageal wall itself, since the esophagus is lined by ciliated columnar epithelium at one time in its embryological development. The paraesophageal cyst lined by ciliated columnar epithelium should be differentiated from paraesophageal cysts which are lined by mucous membrane similar to that of the stomach or small intestine. Paraesophageal cysts lined by gastric mucosa may

secrete pepsin and acid which result in ulceration and even erosion of adjacent structures. Examination of the contents of the cyst may aid in the differential diagnosis of a paraesophageal bronchiogenic cyst from a thoracic gastric cyst. Both types can bear a similar relationship to the esophagus, and it would not always seem possible to make a differential diagnosis on the basis of clinical and roentgen findings alone. An analysis of the literature suggests that the thoracic gastric cysts are far more likely to lead to symptoms and surgical intervention in early life than the paraesophageal bronchiogenic group. Schwartz and Williams[35] collected ten cases from the literature and added two of their own. Half of the cases of thoracic gastric cysts caused symptoms or death within the first year of life, and only two of the 12 cases were over four years of age at the time of operation or autopsy. Probably the gastric mucosa secretes more rapidly and leads to more rapid enlargement of the cyst, with secondary compression of the lung. The mediastinal cysts of enteric origin usually project from the right side of the mediastinum, and although the bronchiogenic cysts are also more common on the right side, there is not the overwhelming predilection for the right side as is seen with those of enteric origin. No obvious sex predominance has been noted in either group. Robbins[32] reported a case of a small bronchiogenic cyst which was located in the wall of the esophagus and appeared as an intramural, extramucosal lesion. The mucosal folds of the esophagus were preserved over the lesion and the mass was noted to move up and down with respiration and upward during the act of swallowing. The motion of the mass was similar to that of the esophagus. At operation the cyst was removed from the wall of the esophagus, and histopathologic examination showed respiratory ciliated epithelium, smooth muscle and cartilage in the wall of the cyst.

TREATMENT

In a monograph on mediastinal tumors published in 1940, Heuer and Andrus[16] state that they had found in the literature 25 cases of mediastinal cysts. This group of cases apparently includes both bronchiogenic and esophageal cysts. In 12 instances an operation had been undertaken for possible removal of the cyst, but the tumor was excised in only 8 of these cases. In 5 instances the cyst was extirpated at the first operation and in 3 its removal followed drainage and marsupialization. Operation in the remaining 4 patients consisted either of drainage of the cyst or its partial removal. All but one of the reported cases in whom complete removal of the cyst was possible recovered, but one of them had a persistent bronchial fistula. The authors also reported a personal case in which recovery followed excision of the cyst. During the past few years several reports on the surgical extirpation of bronchiogenic mediastinal cysts have appeared. The recent increase in routine roentgenographic examination has been responsible for the finding of a large number of these cases in which the patient was asymptomatic. Bronchiogenic cysts of the mediastinum were considered rare a few years ago, but are known today to be one of the most common tumors of the mediastinum.

Adams and Thornton[1] reported three cases of bronchiogenic cyst of the mediastinum treated successfully by surgery. Brown and Robbins[5] analyzed 12 cases of mediastinal cyst from the Massachusetts General Hospital. In 6 of the 12 cases the bronchiogenic origin of the cyst may be considered definite on the basis of the histologic appearance of the cyst wall, and in one additional case the gross findings at operation leave little doubt about the diagnosis. In the remaining cases the cyst wall contained no definite bronchiogenic elements.

It is often difficult definitely to diagnose the exact nature of a mediastinal tumor preoperatively. As mediastinal tumors in general should be removed if proper facilities are available and no contraindication exists, a failure to make a correct preoperative diagnosis is not necessarily a great disadvantage. Whereas a considerable number of the bronchiogenic cysts which have been reported in the literature caused few if any clinical symptoms, a sufficiently large percentage cause complications, especially secondary respiratory infections, to warrant surgical removal. Naturally, if the patient's general condition is such as to increase the hazard of surgical intervention, it may be best to merely observe the patient closely if it is considered quite certain that the lesion is a benign cyst.

The author recommends a posterolateral transpleural approach because of the impossibility of ascertaining preoperatively the various attachments of the cyst. A posterolateral approach permits access to all parts of the pleural space and mediastinum. It may not be possible to remove the cyst intact. If the patient has manifested no signs of infection preoperatively, and if the findings at operation do not suggest suppuration, opening of the cyst and evacuation of its contents in the course of its excision would not seem to be hazardous. One wonders whether some of the empyemas following the opening of a bronchiogenic cyst reported in the older surgical literature may not have been due to other causes than opening the cyst.[8] In three of our patients the cyst was opened at operation and there was gross contamination of the pleural space, but no pleural infection ensued. In one patient, (Case 4) it would have been impossible to remove the cyst intact. Before the cyst was opened, it was felt that complete removal might not be possible because of the marked extension of the cyst through the posterior mediastinum to the contralateral side, and because of its close relationship to the pericardium. After the cyst had been opened and evacuated, with collapse of the walls, it was possible to define the limits of the cyst more accurately, and then remove the cyst wall by combined sharp and blunt dissection.

As the cysts are on a congenital basis, the possible presence of other anomalies must be borne in mind. In one patient (Case 4), there was an anomalous pulmonary vein to the right lower lobe which had to be carefully dissected from the cyst wall. The inferior pulmonary vein was many times its normal length and encircled the lower border of the cyst. If this vein had been injured a lobectomy would have had to be performed, in addition to the

removal of the cyst. Similar anomalies of the vascular system may be found in cases of intrapulmonary bronchiogenic cysts.

If complete removal of the cyst wall seems hazardous, it would seem permissible to leave a small portion *in situ*. Adams and Thornton[1] have utilized silver nitrate to destroy the remaining epithelial lining. Incomplete removal of a bronchiogenic cyst is not as likely to lead to complications as partial removal of gastric or dermoid cysts.

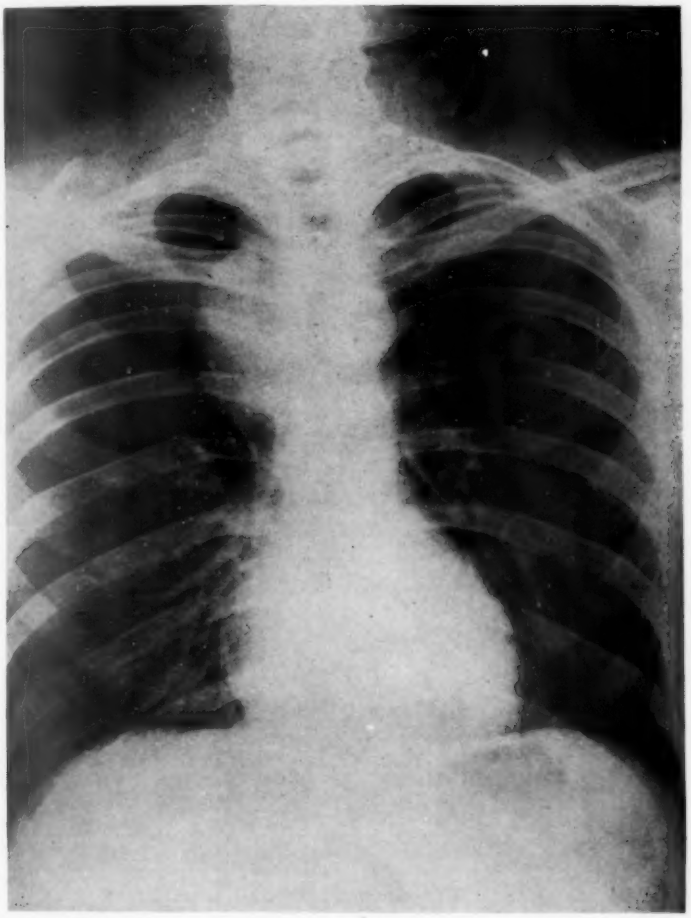


FIG. 3.—Case 1: Paratracheal type of bronchiogenic cyst. Note that upper border of mass does not descend from cervical region in manner commonly seen with substernal extension of a thyroid.

In the paratracheal bronchiogenic cyst, injury to the tracheal wall must be carefully avoided. In our case the cyst had a common wall with the trachea over an area of approximately 2 cm. with a direct continuation of cartilaginous rings from the tracheal wall into the wall of the cyst. These small cartilaginous fragments were removed level with the tracheal wall, but a very small portion

of the cyst wall was actually left adherent to the trachea. In this instance it was not deemed advisable to apply any escharotic to destroy the residual lining because of the danger of denuding the cartilage, and the development of a chondritis. Although there were some recesses in the attachment between cyst and trachea, careful probing failed to reveal any continuity with the lumen of the trachea.

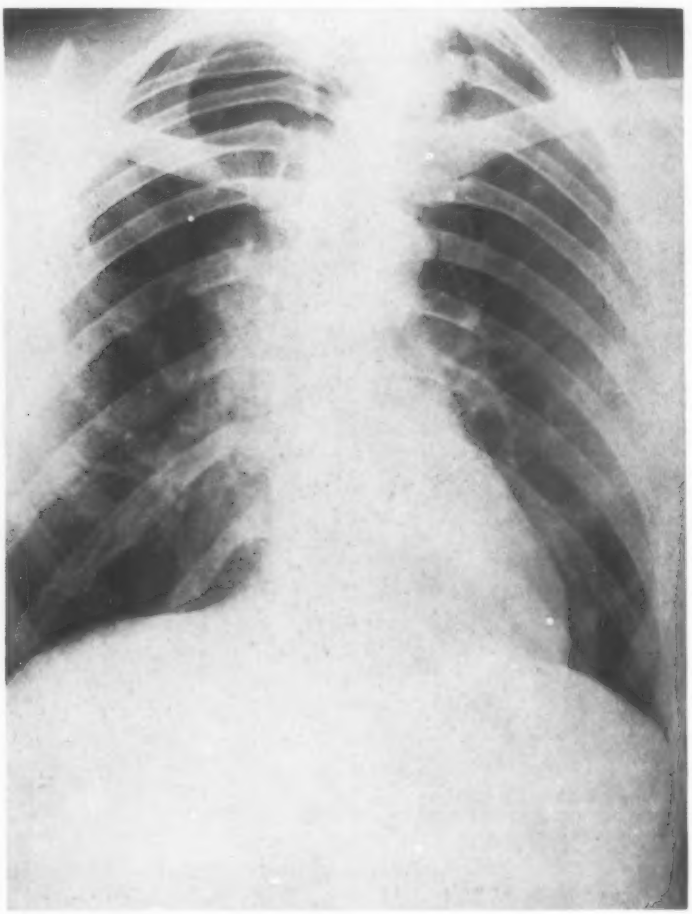


FIG. 4.—Case 2: Bronchiogenic cyst projects laterally above right hilar region. Considerable scoliosis due to hemivertebrae is present.

In operations for mediastinal tumors it is a great advantage to get complete early expansion of the lung. For this reason all the residual air is aspirated from the pleural space through a catheter as soon as the chest wall has been rendered air-tight by closure of the muscles of the thoracic wall. The catheter is then withdrawn as a mattress suture is tied. In some instances closed drainage for a day or two may be desirable.

Most of the serious complications in the small number of surgical procedures that have been carried out for bronchiogenic cysts occurred in the case reports of earlier years and were related to the technic of thoracic surgery in general at that time, rather than to any particular problems encountered in bronchiogenic cysts. Seven of my eight patients had an uneventful postoperative course. One patient, who had mild symptoms of hyperthyroidism prior to operation as well as considerable pulmonary infection from bronchial compression, had a rather stormy postoperative course. At operation the cyst wall was dissected from a large part of the pericardium. This operative manipulation, superimposed on a mild hyperthyroidism, apparently precipi-

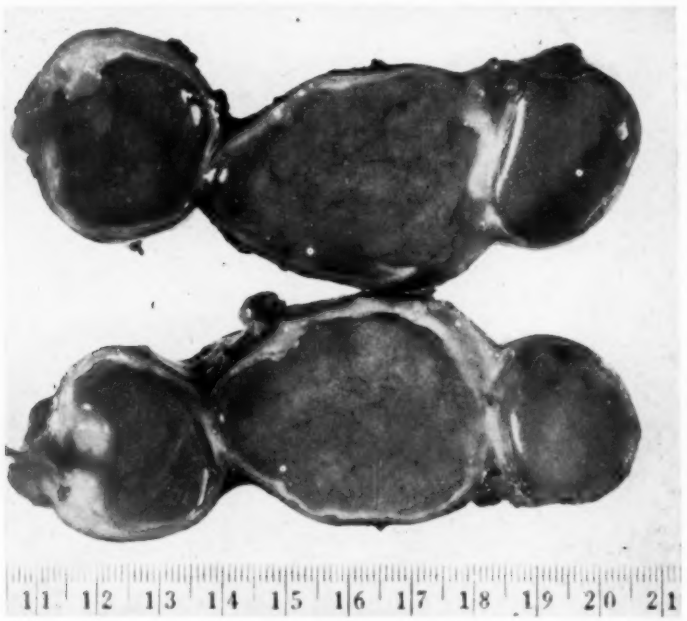


FIG. 5.—Case 2: Cut halves of gross specimen reveal three contiguous cysts.

tated auricular fibrillation and flutter several days postoperatively. The cardiac abnormality rapidly responded to digitalis, and the patient had an otherwise uneventful course.

CASE REPORTS

Case 1.—R. W., #67865, male, age 26 years. Eight years prior to admission a routine chest roentgenogram had shown widening of the upper mediastinal shadow which was interpreted as an enlarged thymus. Subsequent roentgenograms showed little increase in the size of the shadow, until four months before admission to Memorial Hospital when another film showed a definite increase in the size of the mediastinal density. The patient did not complain of any chest pain, cough, expectoration, hemoptysis or dysphagia. There had been no change in weight and his general health was good. Physical examination

was essentially negative. Roentgenograms of the chest in postero-anterior projection (Fig. 3) showed a shadow of considerable density projecting from the right upper mediastinal shadow. The shadow began a short distance above the upper margin of the clavicle and had a slightly irregular but sharply defined border, and extended down to the level of the vena azygos. The outer border of the shadow projected laterally several centimeters beyond the normal mediastinal density. The lateral film showed the density to occupy the upper posterior mediastinal area. There was considerable narrowing of the tracheal air column, especially from the anterior aspect. Roentgenogram of the esophagus revealed no abnormality and no compression by the tumor mass. Angiocardiographic studies showed no obstruction or displacement of the superior vena cava, and indicated that the mass was not an aneurysm. Bronchoscopic examination revealed only

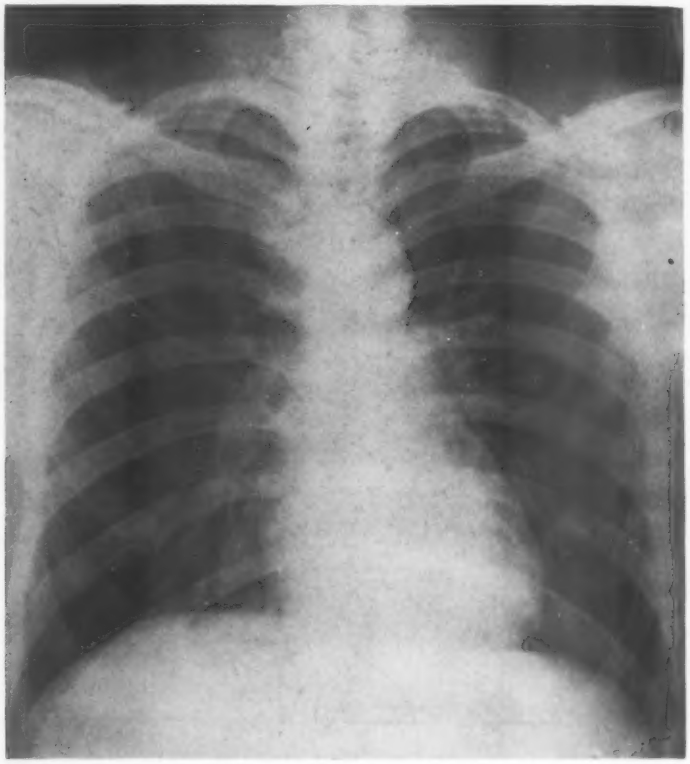
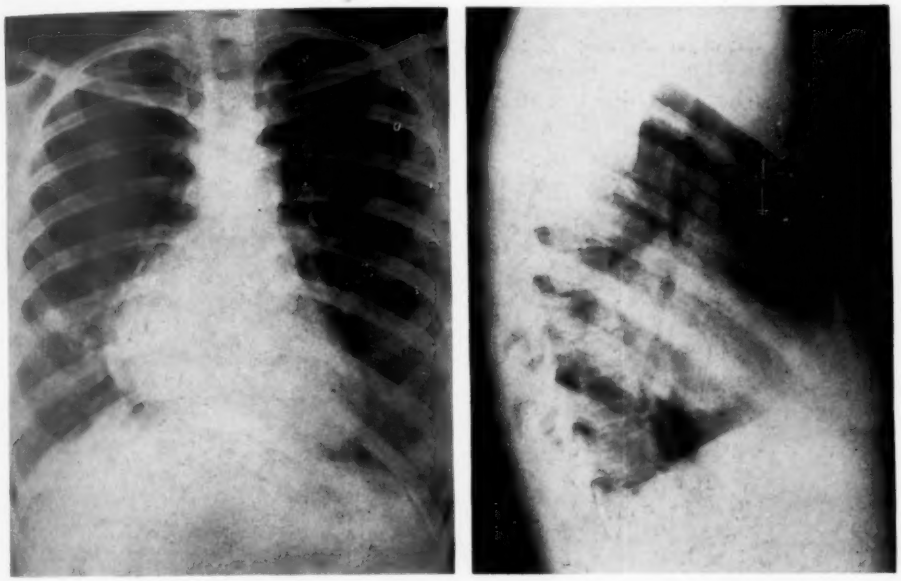


FIG. 6.—Case 3: Small bronchiogenic cyst projects from right side of mediastinum just below level of inner end of clavicle.

compression of the trachea from the right lateral and anterior aspect. The basal metabolic rate was minus 16.

Operation.—In the upper posterior mediastinal region there was bulging of the mediastinal pleura caused by an underlying tumor mass. The mass felt cystic with areas of calcification. The mass extended from the level of the thoracic inlet down to and slightly behind the vena azygos. It extended from the vertebral column to the innominate artery anteriorly. The tumor mass was loosely adherent to the superior vena cava and vena azygos and also rather closely associated with the vagus nerve. The mass, which measured approximately 10 x 6 cm., was adherent to the right lateral wall of the trachea throughout the greater portion of its extent, but was continuous with the tracheal wall

for a distance of about 1 centimeter only. In this area the trachea and cyst wall had a common partition and cartilaginous plaques could be felt extending from the trachea into the base of the cyst. The wall of the cyst was thin and the cyst contained thick, white, non-odorous mucoid material. There was no sign of inflammation. The right lung appeared normal. The entire cyst was removed, with the exception of the small portion which had its wall in common with that of the trachea. No sinus into the trachea could be demonstrated with the aid of a fine probe. Although it was realized that the small portion left attached to the trachea had an epithelial lining, it was deemed inadvisable to apply any caustic because of danger of necrosis of the cartilage of the tracheal wall. The postoperative course was uneventful. Subsequent roentgenograms showed the tracheal air



A B

FIG. 7.—Case 4: A. Postero-anterior roentgenogram reveals area of density merging with right heart border. Note infiltration in right lower lung field.

B. Lateral film shows mass in posterior mediastinum.

column to be of normal dimensions. Three years after operation the patient was asymptomatic.

The surgical specimen consisted of a cystic structure with thin walls containing many irregular pieces of cartilage. Most of the cyst wall measured only 2 mm. in thickness. There were trabeculations in the interior of the cyst (Fig. 1). Microscopic examination revealed that the wall of the cyst resembled the wall of a bronchus. The cyst was lined by ciliated columnar epithelium. The wall contained mucous glands, cartilage, smooth muscle and elastic fibers (Fig. 2).

Case 2.—T. M. #70313, male, age 23 years. Patient had apparently been well until three years previously, when he noted marked weakness and pallor. On examination a very severe anemia was found. A gastrointestinal series was done, but no evidence of peptic ulceration was discovered. A roentgenogram of the chest, however, revealed a mass in the right posterior mediastinal area. Five blood transfusions were given with resultant correction of the anemia. Because of the roentgenologic findings, a diagnosis of probable lymphosarcoma or Hodgkin's disease of the mediastinum was made at that

time, and the patient received radiation therapy. A total of 2,600 roentgens through two ports was given without significant change in the size of the tumor mass. Neither at that time nor in the following three years did the patient complain of any cough, expectoration or chest pain. There had been no dyspnea or difficulty on swallowing. Curvature of the upper thoracic spine had been present since infancy. The patient was referred to Memorial Hospital in 1943, three years after the mass in the chest was first discovered.

Physical examination revealed a well developed and nourished young male. There was rather marked scoliosis of the upper thoracic spine. Physical examination of the heart and lungs was essentially negative. Roentgenograms of the chest showed an irregular, somewhat rounded mass projecting from the right side of the mediastinum (Fig. 4). Lateral film showed the mass to be in the posterior mediastinum close to the esophagus. A barium study revealed no displacement of the esophagus. There was scoliosis of the upper thoracic spine due to hemivertebrae.

Operation.—Located in the posterior mediastinum, beginning at the level of the 7th rib and extending upward beneath the arch of the vena azygos to the thoracic inlet, was

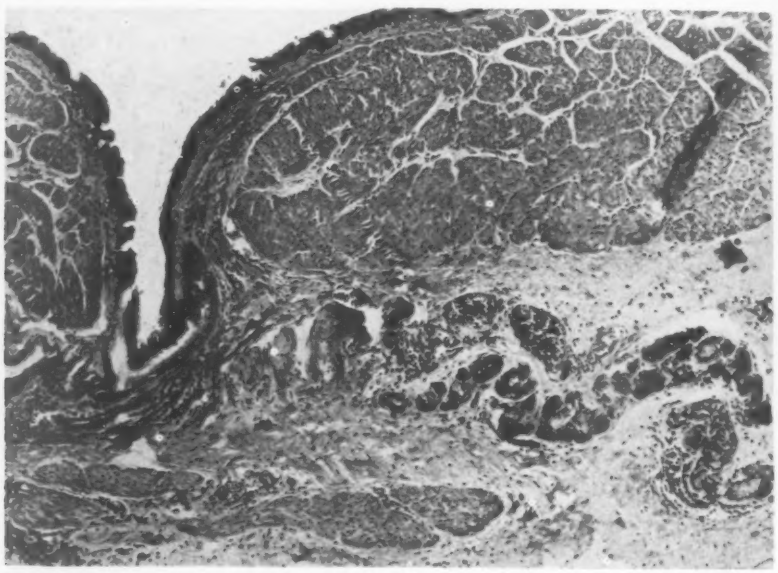


FIG. 8.—Case 4: Photomicrograph of cyst wall shows ciliated columnar epithelium, smooth muscle, and mucous glands.

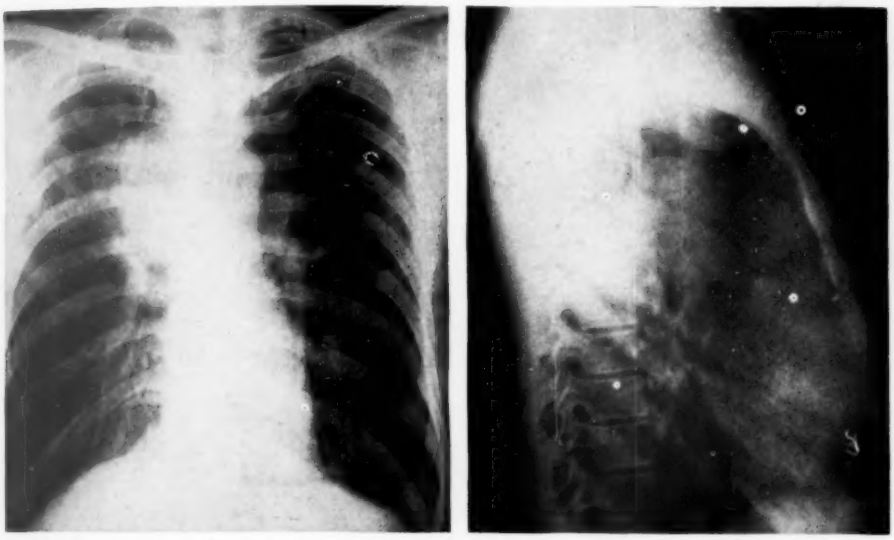
a well encapsulated, lobulated, cystic tumor mass measuring about 10 cm. in its greatest length. The tumor consisted of three apparently separate cystic areas which were attached to each other (Fig. 5). The cystic tumor was not attached to the trachea, but was more closely associated with the right lateral esophageal wall, although there was actually no definite attachment to the esophagus. The tumor was removed after division of the azygos vein. The postoperative course was essentially uneventful. The patient has been well for three years.

The specimen consisted of a cylindroid-shaped tumor measuring 9 cm. in length and 3 cm. in diameter. There were two areas of constriction, so that the specimen had the appearance of being composed of three separate portions. The outer surface showed a fairly smooth capsule. On section, the mass was seen to consist of three definite, apparently non-communicating cysts. All three cysts were filled with semi-viscid, brownish, slightly oily material. The lining of the middle cyst and the upper cyst was smooth and shining, while the lower was finely granular. The cyst wall was firm and white, and

varied from 1 mm. to 1 cm. in thickness. Microscopic examination of the cyst wall showed all the component structures of a bronchial wall except cartilage.

Case 3.—L. F., #70359, male, age 18 years. Two months prior to admission a roentgenogram of the chest had shown a small, rounded density projecting from the right side of the mediastinum. The patient's only complaint was a slight, dry cough which he had had for several years and which he attributed to smoking.

Physical examination revealed a well developed and nourished young male. General examination was essentially negative. No thoracic abnormality could be detected by percussion or auscultation. Roentgenogram of the chest revealed a small, rounded shadow projecting out from the mediastinal density on the right side opposite the level of the arch of the aorta (Fig. 6). The lung fields were clear. On the lateral film the mass could be vaguely outlined in the posterior mediastinum.



A B

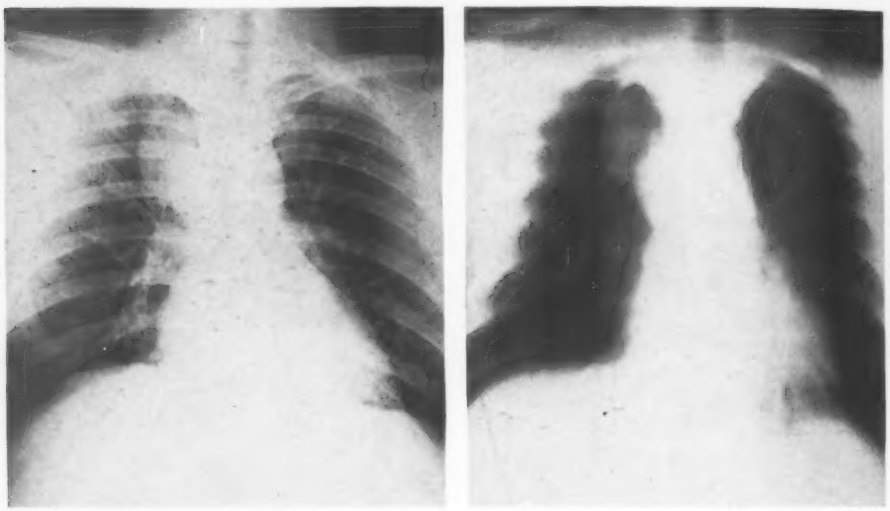
FIG. 9.—Case 5: A. Postero-anterior roentgenogram reveals mass in upper portion of right hilar region. Infiltration of the right upper lobe is evident.
B. Lateral roentgenogram reveals mass in posterior mediastinum.

Operation.—A small multilobular, cystic mass was found just below the vena azygos and attached to the right main bronchus by a small pedicle. The cystic mass was loosely imbedded in the posterior aspect of the right upper lobe, from which it could be separated with ease. The cyst was grayish-white in appearance and the wall was very thin. The entire cystic mass was removed without difficulty. There was no communication between the cyst and the lateral aspect of the right bronchus, to which it was attached. The patient's postoperative course was uneventful. Patient has been well for three years, except for a slight cough.

The specimen consisted of a multilobulated mass 5 x 5 cm. Cut section showed the cyst to contain fluid which was jellied by previous fixation. The cyst was only a few millimeters in thickness. Microscopic examination of the cyst wall showed the various components of a bronchus.

Case 4.—S. B., #70577, female, age 33 years. This patient complained of increasing productive cough of two years' duration with mucopurulent sputum which was frequently blood-tinged. Slight dysphagia had been present for six months. For two months there

had been some pain in the right posterior thoracic region. Physical examination revealed a fairly well developed and nourished negress who did not appear acutely ill. Slight exophthalmos was present and the thyroid was symmetrically enlarged and firm. There was slight tremor of the hands. Examination of the lungs revealed some diminution of breath sounds at the right base posteriorly with occasional rales. The heart was slightly overactive, but no murmurs were heard. Roentgenogram of the chest showed a rounded density projecting from the right lower mediastinal shadow continuous with the density of the right cardiac border. There was scattered infiltration in the right lower lung field (Fig. 7A). On a lateral film the mass was seen to lie in the posterior mediastinum (Fig. 7B). Barium studies showed the esophagus to be displaced toward the left by the mass, but there was no obstruction. The basal metabolic rate was plus 10. It was thought that the patient had a bronchiogenic cyst with compression of the bronchial tree and secondary pulmonary infection with probable bronchiectasis. Bronchoscopy was not done preoperatively because of the patient's refusal following an unsuccessful attempt elsewhere. It was thought the patient had mild hyperthyroidism, but insufficient to contraindicate the thoracic operation as the primary surgical procedure.



A B

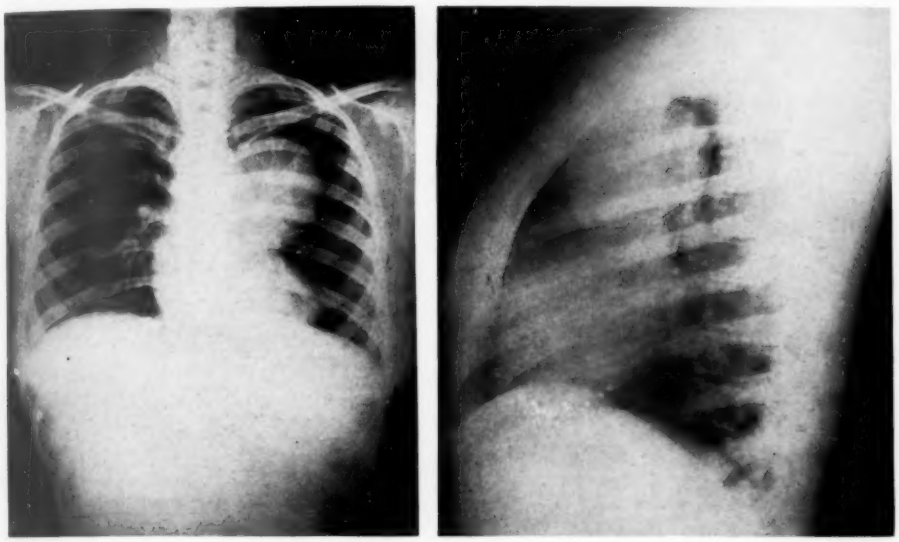
FIG. 10.—Case 6: A. Roentgenogram shows double bronchiogenic cyst projecting from right upper mediastinum.
B. Tomograph shows more clearly the outline of two cysts.

Operation.—Avascular adhesions were present over the lower lobe. A cystic tumor mass occupied the posterior mediastinal area. Considerable difficulty was encountered in freeing the cyst from the inferior pulmonary vein which was elongated and closely associated with the wall of the cyst. During mobilization of the cyst it was ruptured and the contents removed by suction and sponge. The collapsed cyst wall was then dissected free by sharp and blunt dissection. The cyst wall varied from 1 mm. to 4 mm. in thickness. Microscopic examination revealed findings typical of a bronchiogenic cyst (Fig. 8).

Two days postoperatively the patient developed auricular fibrillation which was treated by digitalization and Lugol's solution. Auricular flutter occurred for a brief period. It was thought that the cardiac complication was due to a combination of mild hyperthyroidism and extensive operative manipulation in the region of the auricles. The cardiac rate became normal within a few days. The patient also had considerable cough and expectoration, undoubtedly due to the preoperative pulmonary infection. The

patient was discharged on the 16th postoperative day with the wound well healed. A thyroidectomy was performed later. During the three years since operation the patient has had a productive cough. A resection of the bronchiectatic portion of the lung has been recommended.

Case 5.—J. L., 73999, male, age 35 years. In March, 1943, this patient had a left-sided pleurisy. A respiratory infection developed in January, 1944, following which he had frequent cough with moderate expectoration but no hemoptysis. In March, 1944, he had dyspnea on exertion. A routine chest roentgenogram, taken at an army induction center, revealed a mass in the right paramediastinal area. Patient was admitted to the hospital in May, 1944. Physical examination revealed a somewhat undernourished male who did not appear ill. Physical examination of the chest was essentially negative. Roentgenogram showed a right paramediastinal area of density with infiltration in the adjacent lung



A B

FIG. 11.—Case 7: A. Roentgenogram shows sharply demarcated mass projecting from left hilar region.
B. Lateral roentgenogram shows anterior location of cyst.

(Fig. 9A). On lateral view the density was found to be in the posterior mediastinum (Fig. 9B). There was another small area of infiltration in the right upper lobe separate from the main mass. Fluoroscopy showed no pulsation of the mass. Bronchoscopy was negative.

Operation.—A bronchiogenic cyst of the mediastinum extending into the right upper lobe was found. Evidence of considerable inflammation was present and there was a secondary bronchiectasis of the right upper lobe. The cyst and upper lobe were removed. The postoperative course was essentially uneventful. Within seven months after operation the patient had gained 30 pounds in weight, so that he weighed more than at any previous time. Roentgen-ray showed good expansion of the remaining portion of the right lung.

Pathologic examination showed the wall of the cyst to be largely replaced by granulation tissue, but a respiratory type of epithelium and areas of squamous metaplasia were also found. The excised lobe revealed bronchiectasis.

Case 6.—H. S., Lenox Hill Hospital #108799, male, age 30 years. One year prior

to admission a mass was discovered in the paramediastinal region on a draft board roentgen-ray. Subsequent roentgenographic studies showed a double mass projecting out from the right side of the mediastinum. Patient had no cough or sputum, no chest pain, and his weight was increasing.

Examination revealed a robust male without abnormal physical findings. Fluoroscopy showed a density extending out from the right mediastinal area, which on lateral projection was in the posterior mediastinum. Roentgenograms showed two smooth, oval shadows of increased density, each about 5 cm. in diameter, in the posterior portion of the right upper lobe near the mediastinum (Fig. 10A and 10B). These shadows were in close contact with each other. Their anterior surfaces were in close relation to the right main bronchus. Bronchoscopy showed no evidence of narrowing of the trachea or bronchi but the bronchus of the right upper lobe was somewhat larger than normal.

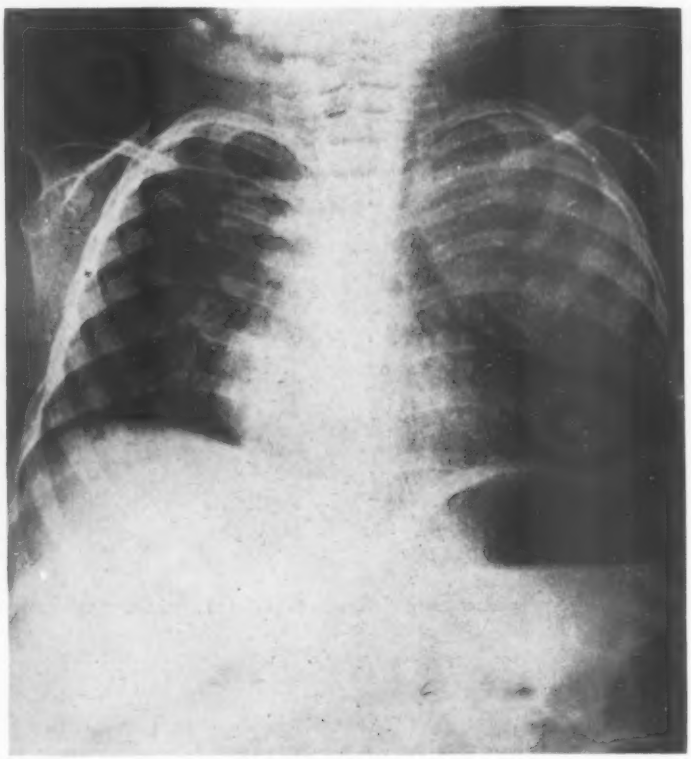


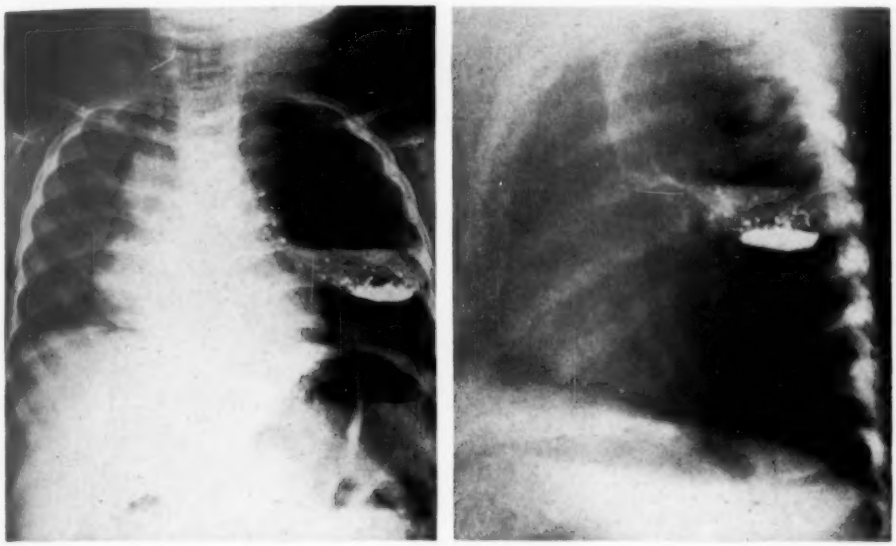
FIG. 12.—Case 8: Large rounded area of density is present in the left upper portion of thorax.

Operation.—A dumb-bell shaped cystic mass was palpable in the hilar region of the right upper lobe. A small portion of this cystic mass could be seen on the posterior aspect of the hilum, but the major portion of the mass was covered by the medial portion of the right upper lobe with which it was intimately associated. The cyst projected for a considerable distance into the pulmonary parenchyma, although there was a line of cleavage between the cyst and the normal pulmonary tissue. The cyst was fairly thin-walled, had cartilaginous plaques within it, and contained non-odorous, greenish-gray, thick material. There was a strand-like thickening in the pulmonary tissue extending from the upper portion of the larger cyst to the apex of the lung, where there was a smaller cyst about 2 cm. in diameter which contained material similar to the larger cyst. Otherwise the pulmonary tissue of the upper lobe appeared relatively normal. The middle

and lower lobes were air-containing and appeared normal. The cysts were excised and the patient's postoperative course was uneventful.

Microscopic examination revealed the inner surface of the cyst to be partially lined with tall, ciliated columnar epithelium supported by a congested stroma which was richly infiltrated with round cells. Embedded in the stroma was an occasional small mucous gland. Portions of the cyst showed marked fibroblastic proliferation, and was densely infiltrated with inflammatory cells, including a few multinucleated giant cells of the foreign body type. In the deeper layers of the wall there was extensive fibrosis and a perivascular focal round cell infiltration.

Case 7.—R. D., Lenox Hill Hospital #109712, female, age 22 years. On a routine chest roentgenogram taken during a physical examination, a mass was found in the left



A B

FIG. 13.—Case 8: A. Roentgenogram taken after aspiration of cyst contents and replacement with air and small amount of lipiodol.

B. Lateral view of the same.

mediastinal region. The patient never had any symptoms referable to the thorax. Physical examination was essentially negative. Roentgenogram of the chest showed a rounded density projecting from the left side of the mediastinum slightly above the hilar region. The mass was of homogeneous density and the margins were smooth (Fig. 11A). A lateral film showed that the mass was located in the anterior mediastinum (Fig. 11B). Preoperative diagnosis was dermoid cyst.

Operation.—A large, rounded, relatively thin-walled cyst occupied the upper anterior aspect of the left pleural space, projecting into it from the mediastinum. The wall of the cyst contained cartilaginous plaques. The attachment of the cyst was near the anterior aspect of the hilum of the left lung in close association with the left main bronchus but not actually attached to the bronchus. Anterior to the attachment of the cyst in the mediastinum there was a congenital defect in the pericardium measuring approximately three centimeters in diameter. This defect was just posterior to the pericardiophrenic vessels and phrenic nerve. Through the defect in the pericardium the main pulmonary artery and the tip of the auricle could be seen. No inflammatory adhesions were present. No other anomalies were noted. The bronchiogenic cyst was excised intact. The defect

in the pericardium was partly closed with the flap of mediastinal pleura which had been dissected from the cyst. The postoperative course was uneventful. There was no accumulation of fluid in the pericardial sac postoperatively, as determined by roentgenograms.

Cut section of the excised cyst revealed a relatively thin-walled sac filled with brownish, thick, mucilaginous material. Several trabeculae were present within the cyst.

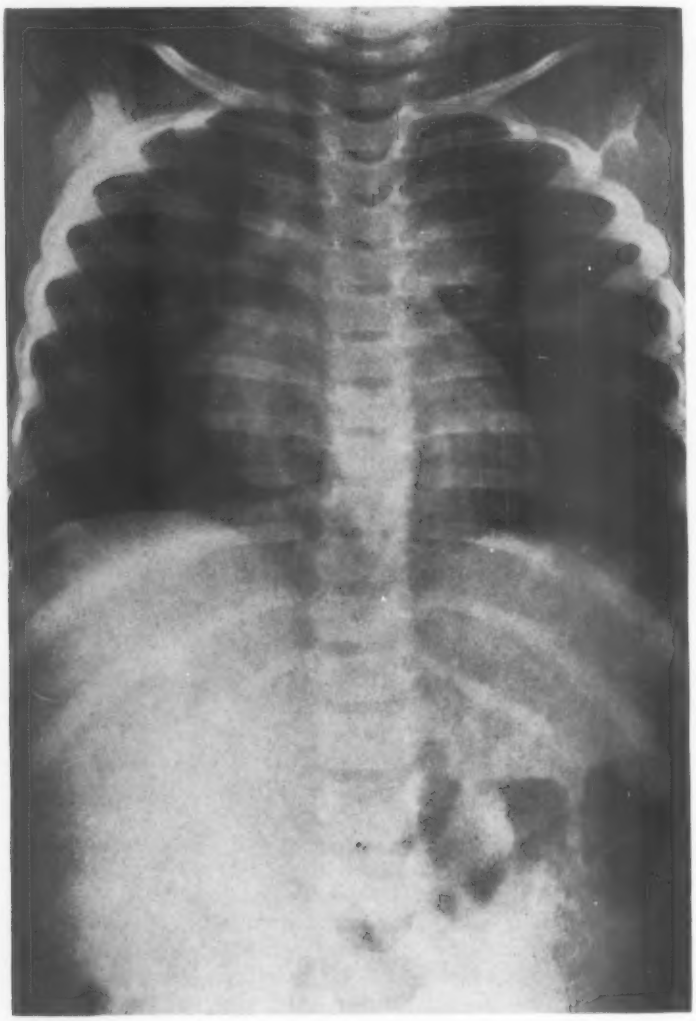


FIG. 14.—Case 8: Roentgenogram after surgical removal of the bronchiogenic cyst. Note well expanded left lung.

Microscopic examination revealed a lining of ciliated columnar epithelium. The underlying stroma contained mucous glands, fragments of cartilage, bundles of smooth muscle and islands of lymphoid tissue.

Case 8.—J. J., female, age nine months. This baby was admitted to Kings County Hospital because of dyspnea and bouts of cyanosis since birth and frequent respiratory infections. A diagnosis of unresolved pneumonia of the left upper lobe had been made. Chest roentgenogram, however, revealed a shadow suggesting a large cyst (Fig. 12). A

needle was introduced into this region and thick, gelatinous, yellow material, which was sterile on culture, was aspirated. At a second aspiration of the cyst some air and a small amount of lipiodol were introduced and further roentgen-rays taken. (Fig. 13A and 13B). A diagnosis of bronchiogenic cyst was made.

Operation.—A large, thin-walled cyst which was attached by a small pedicle to the mediastinum near the anterior end of the interlobar fissure close to the phrenic nerve was found. A structure like a small bronchus could be felt in the mediastinal pedicle of this cyst, but this ended blindly and was not in close association with the remainder of the tracheobronchial tree. The cyst was excised and the postoperative course was uneventful. Microscopic examination revealed the characteristic findings of a bronchiogenic cyst. The lung expanded well after operation (Fig. 14).

SUMMARY

Bronchiogenic cysts of the mediastinum result from the faulty development of elements of the primitive foregut. The cyst wall resembles that of the bronchus. The symptomatology depends chiefly on the size and location of the cyst. The various clinical pictures associated with the lesion are discussed in relation to the site of the cyst. Bronchiogenic cysts are a common type of mediastinal tumor. Surgical excision is usually indicated, although a considerable percentage of the patients are asymptomatic when the lesion is first discovered on a roentgenogram. Eight illustrative cases are reported.

BIBLIOGRAPHY

1 Adams, W. E., and T. F. Thornton: Bronchiogenic Cysts of the Mediastinum. J. Thoracic Surg., **12**: 503, 1043.

2 Alford, J. E.: Congenital Bronchogenic Cyst of the Mediastinum. J. Pediat., **11**: 550, 1937.

3 Altmann, F.: Beitrage zur Lehre von den Lungenmissbildungen. Beitr. z. path. Anat. u. z. allg. Path., **82**: 199, 1929.

4 Blackader, A. D., and D. J. Evans: A Case of Mediastinal Cyst Producing Compression of the Trachea Ending Fatally in an Infant of Nine Months. Arch. Pediat., **28**: 194, 1911.

5 Brown, R. K., and L. L. Robbins: The Diagnosis and Treatment of Bronchiogenic Cysts of the Lung. J. Thoracic Surg., **13**: 84, 1944.

6 Carlson, H. A.: Congenital Cysts of the Mediastinum. J. Thoracic Surg., **12**: 376, 1943.

7 Chiari, H.: Ueber einen neuen Typus von Missbildung an der Trachea des Menschen. Beitr. z. path. Anat. u.z. allg. Path., **5**: 329, 1889.

8 Ehlers, H. W. E.: Zur Kenntnis der intrathoracalen Flimmerepithelzysten. Deutsche Ztschr. f. Chir., **213**: 189, 1929.

9 Fletscher, M.: Congenital Cyst of the Mediastinum. Tr. Path. Soc. London, **48**: 249, 1897.

10 Flint, J. M.: The Development of the Lungs. Am. J. Anat., **6**: 1, 1906.

11 Friedlander, S. O., and P. W. Gebauer: Disease of Aberrant Intrathoracic Lung Tissue. J. Thoracic Surg., **8**: 581, 1939.

12 Gold, E.: Uber Bronchuscysten und deren Entstehung. Beitr. z. path. Anat. u. .z. allg. Path., **68**: 278, 1921.

13 Guillery, H.: Eine in die Wirbelsaule eingewachsene mediastinale Zyste (Vorderdarmzyste). Centralbl. f. allg. Path. u. path. Anat., **69**: 49, 1937.

14 Harrington, S. W.: Surgical Treatment of Intrathoracic Tumors. Arch. Surg., **19**: 1679, 1929.

15 ————: Surgical Treatment of Mediastinal Tumors; removal of cystic azygos lobe from posterior mediastinum. Ann. Surg., **96**: 843, 1932.

16 Heuer, G., and W. Andrus: The Surgery of Mediastinal Tumors (A Monograph). Am. J. Surg., **50**: 143, 1940.
17 Joel, J.: Ein Teratom auf der Arteria pulmonalis innerhalb des Herzbeutels. Virchows Arch. f. path. Anat., **122**: 381, 1890.
18 Johnston, L. M.: Congenital Ciliated Columnar Epithelial Cysts of the Mediastinum. Am. J. Dis. Child., **56**: 313, 1938.
19 Kern, W.: Beitrage zur Pathologie des Oesophagus. Virchows Arch. f. path. Anat., **201**: 135, 1910.
20 Ladd, W. E., and H. W. Scott, Jr.: Esophageal Duplications or Mediastinal Cysts of Enteric Origin. Surgery, **16**: 815, 1944.
21 Lambert, A. V. S.: Etiology of Thin-Walled Thoracic Cysts. J. Thoracic Surg., **10**: 1, 1940.
22 Maier, H. C., and C. Haight: Large Infected Solitary Pulmonary Cysts Simulating Empyema. J. Thoracic Surg., **9**: 471, 1940.
23 Maier, H. C.: Pulmonary Cysts. Am. J. Surg., **54**: 68, 1941.
24 Melchior, E.: Zur Kenntnis der kongenitalen tracheobronchialen Cysten der Lunge. Zentralbl. f. Chir., **56**: 2626, 1929.
25 Mixter, C. J., and S. H. Clifford: Congenital Mediastinal Cysts of Gastrogenic and Bronchogenic Origin. Ann. Surg., **90**: 714, 1929.
26 Mohr, R.: Zur Kenntnis der Cysten des Mundbodens und des Oesophagus. Beitr. z. path. Anat. u. z. allg. Path., **45**: 325, 1909.
27 Möller, P.: Congenital Thoracic Cysts and Lung Deformities in the Roentgen Picture. Acta radiol., **9**: 460, 1928.
28 Nössen, H.: Tod under dem Bilde der Lungenembolie durch Zyste im Perikard. Deutsche med. Wchnschr., **51**: 1150, 1925.
29 Pugh, D. G.: Pulmonary Cysts (A Review with Bibliography). Am. J. M. Sc., **208**: 673, 1944.
30 Rau, F.: Flimmerepithelzyste des Oesophagus. Virchows Arch. f. path. Anat., **153**: 25, 1898.
31 Rizzi, I.: Le Cisti Endotoraciche; a proposito di una cisti bronchiale del mediastino. Arch. Ital. di anat. e istol. path., **8**: 689, 1937–1938.
32 Robbins, L. L.: The Roentgenologic Appearance of "Bronchiogenic" Cysts. Am. J. Roentgenol., **50**: 321, 1943.
33 Rusby, N., and T. H. Sellors: Congenital Deficiency of the Pericardium Associated with a Bronchogenic Cyst. Brit. J. Surg., **32**: 357, 1945.
34 Schneider, P.: Morphologie der Missbildungen des Menschen. Die Missbildungen der Atmungsorgane, Jena 1912 (Gustav Fischer).
35 Schwartz, H., and C. S. Williams: Thoracic Gastric Cysts. J. Thoracic Surg., **12**: 117, 1942.
36 Seybold, W. B., and O. T. Claggett: Presternal Cysts. J. Thoracic Surg., **14**: 217, 1945.
37 Smith, R. E.: A Case of Mediastinal Dermoid Cyst in an Infant. Guy's Hospital Report, **80**: 466, 1930.
38 Tresp: Flimmerepithelcyste der hinteren Osophaguswand. Arb. aus dem Pathologie Inst. Posen herausgeg. von Lubarsch, Weisbaden, 1901.
39 v.Wyss, H.: Zur Kenntnis der heterologen Flimmercysten. Virchows Arch. f. path. Anat., **51**: 144, 1870.
40 Walzel, P.: Lungentumoren mit Einschluss von auf die Lunge uber greifenden Mediastinaltumoren und Thoraxwandgeschwulsten. Beitr. z. klin. Chir., **158**: 645, 1933.
41 Westenryk, V.: Zur Casuistik der Mediastinal Cysten. Prag. med. Wochnschr., **25**: 373, 1900.
42 Zahn, F. W.: Uber mit Flimmer-epithelien ausgekleidete Cysten des Osophagus. Virchows Arch. f. path. Anat., **143**: 171, 1896.

MID-LEG AMPUTATIONS FOR GANGRENE IN THE DIABETIC

SAMUEL SILBERT, M.D.

NEW YORK, N. Y

FROM THE SURGICAL SERVICE, MONTEFIORE HOSPITAL, NEW YORK

DIABETES appears to be a progressive disease, in spite of the apparently satisfactory control of the carbohydrate metabolism by insulin. Banting's brilliant discovery opened a new era in the treatment of diabetes, and patients with this disease were led to believe that they could look forward to a normal, healthy adult life if they continued to observe care in diet and insulin administration. Among the profession at large an attitude of complacency has developed, and it is widely assumed that the diabetic is well controlled when the blood sugar level is approximately normal and there is no glycosuria. Students of this disease, however, are agreed that control of the carbohydrate metabolism can prevent coma and relieve symptoms, but that the more profound disturbances resulting from diabetes are not corrected by the use of insulin, and that progressive deterioration of the vascular system of the entire body takes place, resulting in albuminuria, hypertension, retinal changes, cardiac and peripheral vascular disease.

As a surgeon familiar with the frequent necessity for amputation in the pre-insulin days, I anticipated that the universal use of insulin would be reflected in a gradual decrease in the incidence of gangrene and amputations in patients with diabetes. It is an outstanding fact that such has not been the case, and it is unhappily accurate to state that because of insulin, many more diabetics live long enough to require amputations of one or both legs. Furthermore, by the time a diabetic has reached the point where he requires amputation of a leg for gangrene, his life has nearly run its course, and he will be among the select few if he is alive five years later. If alive, it is probable that loss of the second leg will have been necessary.

Contributing to the present unsatisfactory picture of diabetes is the high mortality which results from thigh amputations for gangrene. With very few exceptions, the prevailing death rate ranges from 25 per cent to 50 per cent. Table I presents the reported mortality for this procedure in a group of metropolitan hospitals. There were 547 deaths, or 44 per cent, in 1,242 cases of mid-thigh amputations in diabetics. Excluded from this table are the statistics of the Deaconess Hospital in Boston, because of the unique record of this institution. Joslin states that "major amputations numbering 767 between 1932 through 1942, showed a mortality of 12.3 per cent."[18] This unusually low mortality may be due to special factors such as inclusion of many patients of the affluent class, and to the practice of amputation for very early lesions of the extremities. It is apparent that such a low mortality is not characteristic of the country at large, particularly in the large municipal hospitals.

Since the technical procedure of a thigh amputation is simple and requires

no great skill, the prevailing high mortality indicates that this procedure is too severe for the average patient with diabetes. It is readily apparent why this is so. The patient with diabetic gangrene is usually a poor operative risk. He is past middle age, and often has complicating arteriosclerotic cardiorenal or cerebrovascular disease. His vitality may have been reduced by prolonged suffering and by absorption of toxic products from his gangrenous or infected

TABLE I.—*Mortality Following Thigh Amputations for Gangrene in the Diabetic*

	Cases	Deaths	Period	Per Cent Mortality
Montefiore (1)	17	10	1932—1936	59
Mount Sinai (1)	68	26	1926—1936	38
Morrisania (2)	45	27	1931—1935	60
Bellevue, 1st division (a)	40	21	1931—1935	52
Bellevue, 2nd division (a)	35	22	1931—1935	63
Bellevue, 3rd division (a)	70	26	1931—1935	37
Bellevue, 4th division (a)	24	18	1931—1935	75
Lenox Hill (3)	13	5	1935—1939	38
St. Luke's (4)	25	9	1934—1938	36
New York (5)	31	9	1932—1940	29
Roosevelt (6)	12	3	1935—1939	25
Mary Immaculate (Jamaica) (7)	24	12	1930—1935	50
Israel Zion (8)	99	32	1934—1943	32
Kings County (9)	73	34	1936—1941	46
Massachusetts General (10)	36	12	1916—1926	33
Philadelphia General (11)	130	73	1926—1933	56
Philadelphia General (12)	127	61	1937—1939	48
Philadelphia Episcopal (13)	56	27	1926—1935	48
New Orleans Charity (14)	114 (b)	48	1929—1937	42
Indianapolis City (15)	78 (c)	31	1930—1938	40
Rochester University (16)	106 (c)	34	(Not stated)	32
Nebraska University (17)	19	7	1932—1942	37
Total	1242	547		44

a. Reported at N. Y. Academy of Medicine, May 11, 1937.
b. Diabetic and non-diabetic arteriosclerosis.
c. Thigh and leg amputations combined.

foot. To relieve pain he has received considerable quantities of narcotic drugs. It has been difficult to control the carbohydrate metabolism properly because of the diminished effectiveness of insulin in the presence of infection and gangrene. Such a patient should be subjected to as little operative trauma as possible. The operative procedure should be brief, profound anesthesia should be avoided, and amputation should be carried out as far distally as possible.

It has been taught for many years that amputations should be done through the thigh in order to insure adequate circulation for healing. This widespread belief has been proven incorrect by numerous surgeons in the past few years. McKittrick,[19, 20] Beverly Smith,[21] Maes,[22] Crossan,[13] Bickel,[23] and others, have advocated amputations below the knee, and have reported good

results following this procedure. My own experience, likewise, indicates that amputations can be performed safely below the knee in diabetic patients, even when the popliteal artery is closed and oscillometric readings indicate a seriously deficient circulation.

CLINICAL MATERIAL

The present series consists of 127 instances of extensive gangrene with or without infection in diabetic patients treated by mid-leg amputation during the years from 1940 to 1946. Fifty-eight of these patients were ward cases at the Montefiore Hospital, and the other 69 were seen in private practice. In addition, 24 non-diabetic patients with arteriosclerotic gangrene have been similarly treated, making a total of 151 amputations. Advanced impairment of circulation was present in all cases. In most of them the popliteal pulse was absent, and in some even the femoral pulse could not be felt. Only rarely during this period was a thigh amputation done in a patient with diabetes, and only for exceptional reasons, such as marked contracture of the knee joint or unusually poor condition of the tissues of the leg. The majority of the operations were done by the writer, but many were done by colleagues or house staff surgeons under his supervision. It is important to point out that patients admitted to the wards of Montefiore Hospital are frequently in rather poor condition due to age, poor economic status, and prolonged chronic illness. Many of them have passed through other hospitals, and are transferred to Montefiore as chronic invalids. In general, it may be stated that in New York, recommendations for amputations are not readily accepted by most patients, and usually consent is finally obtained only when extensive gangrene and infection have developed. This is a factor which has a considerable bearing on mortality figures. An attempt has been made to grade the general condition of each patient depending upon age, toxicity, cardiac status, presence of hypertension, and degree of peripheral vascular involvement. Table II summarizes this information, and contrasts the mortality in the different groups.

TABLE II.—*Mortality in Diabetic Gangrene Depending Upon General Condition of Patients**

Group	Condition	Number of Patients	Operative Deaths	Mortality Per Cent
A.....	Good	8	0	0
B.....	Fair	34	1	3
C.....	Poor	50	5	10
D.....	Precarious	35	6	17
Totals.....		127	12	9.4

* Division of diabetic patients into groups based upon age, toxicity, cardiac status, hypertension and degree of peripheral vascular impairment.

TECHNIC OF OPERATION

The patient is placed on the table face down, as this position permits flexion of the leg, and makes the operation much easier. Spinal anesthesia, using less than 100 mg. of novocaine, or light general anesthesia is employed. A tourniquet is not used. A circular incision is made through the skin and fascia at a level 8 inches below the patella. Flaps of any kind are avoided. The muscles are divided at the level of the retracted skin. As soon as the superficial layer of calf muscles is sectioned the posterior tibial vessels and nerve are exposed, lying on the deep layer of muscles. The vessels and nerve are ligated and divided and the nerve is injected with alcohol. The leg is then flexed and the anterior tibial group of muscles is sectioned, exposing the anterior tibial vessels and nerve near the interosseous membrane. These structures are then ligated and cut, thus controlling the major sources of bleeding. The leg is again placed horizontal and the deep layer of muscles on the posterior surface is sectioned. The muscles are then separated for a few inches from the bones and the bones are sawed through, the tibia about one inch and the fibula about two inches above the level of the skin incision. Periosteum and bone are cut at the same level. The anterior edge of the tibia is then beveled by an oblique saw cut. Any muscle that has been damaged during the procedure is trimmed away, and careful hemostasis obtained. The wound is thoroughly irrigated with sterile water. The wound is left wide open and is dressed with a combination of paraffin mesh and vaseline gauze. A posterior molded plaster splint is applied and the dressing is not changed for a week. Thereafter the wound is dressed with cod liver oil ointment and the dressing is changed every third day until the stump is healed. There is frequently considerable secretion from the wide open wound and usually some superficial slough of damaged tissue for the first two or three weeks. Then the wound gradually becomes a clean, granulating surface in the center of a rapidly contracting scar. Infections of the stump or other complications are rare. The postoperative course is usually smooth and painless.

It is important to continue the use of the posterior molded splint until the wound is well on the way to healing. Contracture at the knee takes place readily unless a splint is used, and such a contracture is difficult to overcome. In the first few cases a tourniquet was used around the thigh to expedite the operative procedure. Three patients developed gangrene of the leg stump and required secondary thigh amputations. The use of a tourniquet was therefore abandoned. Since then more than 120 consecutive cases have had mid-leg guillotine amputations without the use of a tourniquet, and in only three of these has a higher amputation been necessary. Healing in all patients has been surprisingly good. In a few, minimal necrosis of the skin margin has developed, but this has not interfered with a satisfactory end result.

The process of healing presents some surprises. As soon as a rim of scar tissue forms at the periphery of the wound, contraction of the scar tissue begins and gradually pulls the skin down over the end of the stump (Fig. 1 and 2). This process continues until healing is complete, and the final scar is

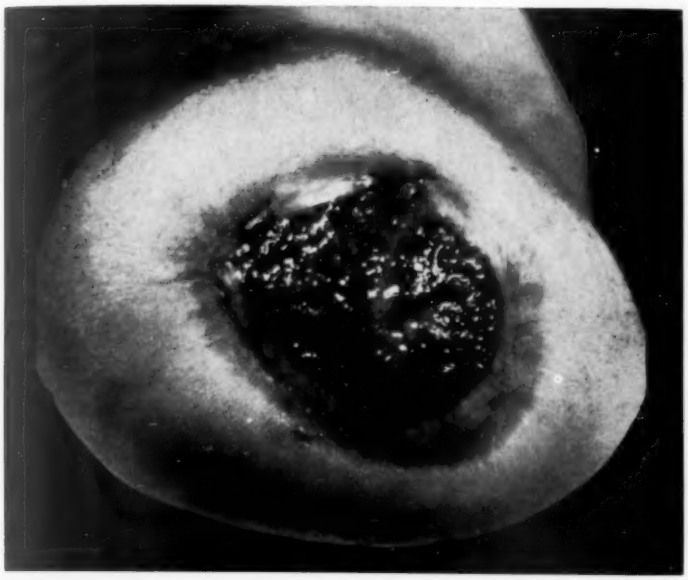


FIG. 1.—Early stage of healing. Note skin drawn over end of stump by contracting scar tissue.

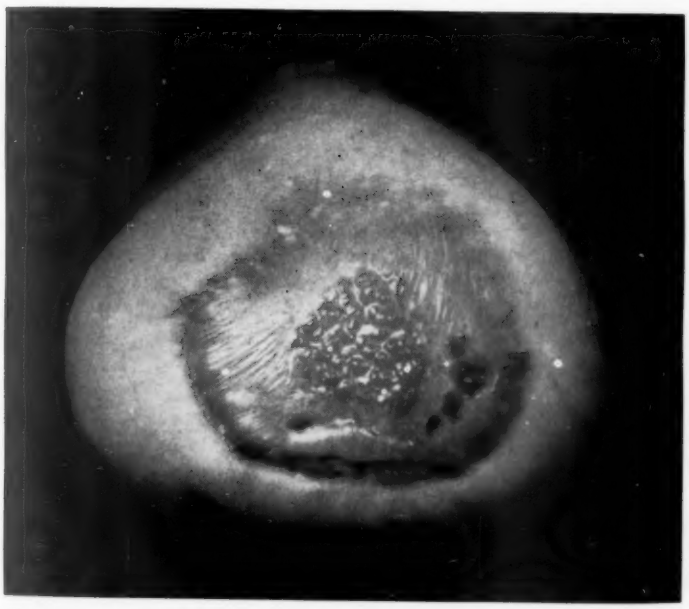


FIG. 2.—Later stage of healing. Small granulating wound in center of contracting scar.

frequently so small that it can be covered with a 25 cent silver coin (Fig. 3 and 4). *It is not necessary to apply any form of traction to the stump to accomplish this result.* The pull of the contracting scar tissue is sufficient. Complete healing usually requires from 10 to 12 weeks. Patients are allowed out of bed the day after operation in most cases, and can leave the hospital on crutches four to six weeks after operation.

RESULTS

FIG. 3.—Well-healed stump. Note relatively small area of scar to circumference of stump.

Every death which occurred before the stump was healed is regarded as an operative mortality, even though the cause of death was entirely unrelated to the surgical procedure. In the entire group of 127 diabetic patients there were 12 deaths, a mortality of 9.4 per cent. In the ward group of 58 patients there were 7 deaths (12.1 per cent); in the private group of 69 patients there were 5 deaths (7.2 per cent). The significance of these figures is clear. It is not necessary to have a high mortality in amputations for gangrene in the diabetic. When the severity of the operation is reduced to the limited endurance of the patient, this mortality can be reduced to less than 10 per cent.

The non-diabetic group of arteriosclerotic gangrene is similar in age and general condition to the diabetic group, and for the sake of completeness may be included with the latter. In this group of 24 cases there were 2 deaths. Thus, in the total number of 151 patients treated by mid-leg amputation, there were 14 deaths, a mortality of 9.3 per cent.

Certain other advantages of the low amputation should be stressed. The use of an artificial limb is greatly facilitated if the patient retains his knee joint and about six inches of his leg (Fig. 5). Such patients are frequently able to walk without the use of a cane or crutches, and with scarcely any perceptible limp. On the contrary, when amputations are done through the thigh, experience has shown that almost none of the women and only about half of the men ever accustom themselves to the use of an artificial leg. Many of the 151 patients in the present series, including one man and one woman over 80 years of age, have been fitted with and are wearing artificial legs. I know of no instance where a well healed stump has broken down and required further surgery.

It is worth recording that none of the below-knee stumps have been per-

sistently painful. Pain in *thigh* stumps is common, and is one of the most distressing complications of amputations of the lower extremities. It is frequently not relieved by injections or reoperations on the sciatic nerve or sympathetic nervous system, and chordotomy and excision of parts of the cerebral cortex have been employed in an effort to control it. I do not know why leg stumps are painless, but this is one of the great advantages of the low amputation.

Are there any contra-indications to the mid-leg amputation? In my opinion there is only one group of cases that is not suitable for this procedure. These are the patients who have had a recent thrombosis of the

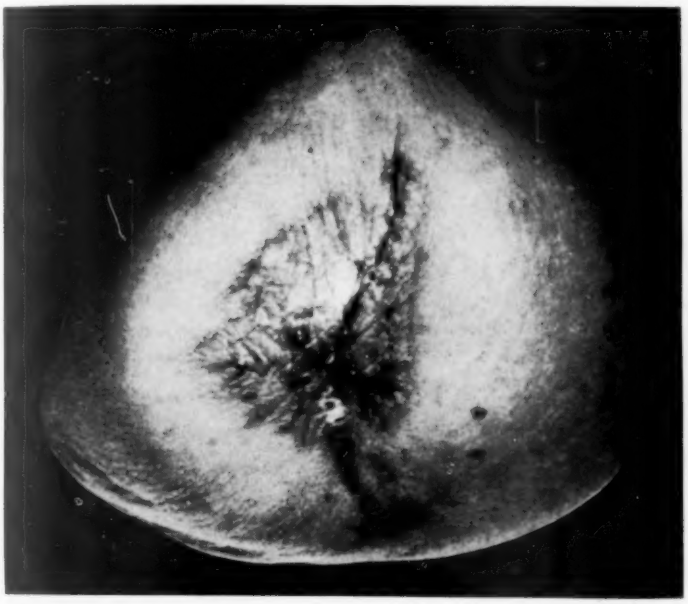


FIG. 4.—Characteristic small scar at end of well-healed stump.

femoral artery, and gangrene develops within a few weeks of onset. Such cases are readily identified, as they give a history of abrupt onset of pain without preceding intermittent claudication. Unless there has been an interval of at least 3 months after the arterial occlusion, there has not been time for an adequate collateral circulation to develop to the mid leg, and it is not safe to amputate below the knee. It has been my experience that such cases are relatively few.

PRIMARY CLOSURE OF MID LEG AMPUTATION

In the first 100 diabetic cases treated by guillotine amputation without any attempt at closure, there were 8 deaths (8 per cent). Having obtained this basic mortality figure, some cases were selected in the second hundred for

FIG. 5.—Well-healed stump after guillotine amputation. Preservation of knee joint and upper part of leg improves ability to walk with artificial leg.

primary closure following amputation at this level. With this modification healing is complete in 2 to 3 weeks. There are three dangers in primary closure: infection, tension on tissues causing impairment of circulation, and inadequate circulation at the operative level causing localized necrosis of tissue. In cases treated by primary closure an attempt to prevent infection was made by treating each case for one week before and one week after operation with injections of penicillin. Care was used to avoid tension on the suture line and few sutures were used. In 6 selected cases in the diabetic group treated in this manner, 4 healed by primary union, and 2 had a narrow area of gangrene along the suture line which delayed healing but did not prevent an ultimately satisfactory result. In addition, 3 cases in the non-diabetic group, closed in the same way, healed by primary union.

While the results in this small group of 9 cases has been all that could be expected, it is still a question whether it is desirable to take the additional risk of primary closure to save a few weeks of healing time. Most of the diabetic patients who require amputation are beyond 60 years of age and are no longer engaged in active work. The granulating wound resulting from a guillotine amputation left entirely open is not painful and does not require hospitalization. Visits on crutches to the surgeon's office twice a week are sufficient for dressing the stump. The advisability of primary closure will depend on further experience with this modification.

NECESSITY FOR AMPUTATION OF SECOND LEG

The number of patients with diabetes who require amputation of the second leg has not been determined in a large series of patients. The American Diabetes Association has no data on this point. Joslin[18] stated that of 100 patients, 39 subsequently required amputation of the second leg. In the present series of 127 patients, 20 required amputation of the second leg during the known period of observation, and the follow-up is incomplete. When patients who have been followed for only a short period are excluded, more significant figures are obtained. For example, among the diabetic patients

seen in my private practice, there are 49 with one amputation who have been followed for over 3 years. Among these, 21 or 43 per cent, have required amputation of the second leg. This figure closely approximates that of Joslin cited above. Thus it appears that about 40 per cent of diabetic patients who survive 3 years after the loss of one leg will require amputation of the second leg. More extensive data on this point are desirable, and a significant reduction in the percentage of patients who required amputation of the second leg could be accepted as evidence of the value of the prophylactic treatment used in the interval. Ligation of the femoral vein of the second leg is being done at the time of primary amputation in a series of cases at the Montefiore Hospital to determine if this procedure has any merit as a prophylactic measure. It will take many years to determine this point. Sympathectomy might also be tested in this manner.

DURATION OF LIFE AFTER AMPUTATION OF LEG

It has been stated that the average expectancy of patients with diabetes after amputation of one leg is about 3 to 4 years.[24] Experience with the present series of cases confirms the above statement. Thirty-five (27 per cent) of the 127 cases in this series are known to be dead within 3 years after amputation, and since the follow-up is incomplete, the real figure may well be twice as high. Twenty-four patients of this series are alive and comparatively comfortable 3 to 5 years after amputation. Forty-nine of a series of 117 diabetic patients with amputations seen in my private practice have survived 3 years or more since operation (41 per cent). Thus it appears that only about 40 per cent of diabetic patients will live more than 3 years after amputation of a leg.

SUMMARY AND CONCLUSIONS

Although the discovery and use of insulin has made the diabetic patient more comfortable and has prevented diabetic coma, the more profound disturbances are not corrected and diabetes remains a progressive, degenerative disease.

Amputations for gangrene in the diabetic are more frequent than in pre-insulin days.

Routine mid-thigh amputations result in a high operative mortality, unnecessary loss of function, and many painful amputation stumps.

Mid-leg amputations are advocated because of low operative mortality, preservation of better function, and freedom from stump pain. In 127 diabetic patients subjected to mid-leg amputation there were 12 deaths, a mortality of 9.4 per cent.

Only about 40 per cent of diabetic patients will survive longer than 3 years after amputation of a leg. Of those who survive about 40 per cent will require amputation of the second leg.

BIBLIOGRAPHY

[1] Silbert, Samuel: Conservative Surgical Care of Diabetic Gangrene. J. Mt. Sinai Hosp., **5**: 376, 1938.

[2] Williams, Frederick W., and Thomas J. O'Kane: Mortality in Surgical Diabetes-Criteria and Technique in Extremity Lesions. Five-year study of 496 cases. Surg., Gynec. & Obst., **64**: 956, 1937.

[3] Donehue, F. McG.: Surgical Aspects of Diabetic Gangrene. New York State J. Med., **42**: 511, 1942.

[4] Solley, Fred W.: Diabetic Lower Extremity Amputations. New York State J. Med., **42**: 507, 1942.

[5] Conway, H., and S. C. Meigher: Infection and Gangrene of the Lower Extremity in Diabetes Mellitus. New York State J. Med., **42**: 519, 1942.

[6] Pennoyer, G. P.: Diabetic Gangrene. New York State J. Med., **42**: 513, 1942.

[7] Levin, Charles M., and Frank N. Dealy: The Surgical Diabetic. A five-year survey. Ann. Surg., **102**: 1029, 1935.

[8] Mandelberg, A., and W. Sheinfeld: Diabetic amputations. Am. J. Surg., **71**: 70, 1946.

[9] Al Akl, F. M., A. Singer, and C. B. Roesch: Peripheral Vascular Surgery. Am. J. Surg., **55**: 520, 1942.

[10] Kuhns, J., and P. Wilson: Major Amputations. Analysis and study of end results in 420 cases. Arch. Surg., **16**: 887, 1928.

[11] Eliason, Eldridge L.: Surgery of Diabetic Gangrene. Ann. Surg., **98**: 1, 1933.

[12] Erb, William H.: Conservative Surgery in Diabetic Gangrene. Pennsylvania M. J., **44**: 1131, 1941.

[13] Crossan, Edward T.: Two Stage Amputation for Diabetic Gangrene of Leg. Am. J. Surg., **33**: 18, 1936.

[14] Veal, James R.: Factors in the Mortality Rate of Arteriosclerotic Gangrene. J. A. M. A,. **110**: 785, 1938.

[15] Taylor, F. W.: Arteriosclerotic Gangrene. J. A. M. A., **113**: 1196, 1939.

[16] Pearse, H. E., and H. R. Ziegler: Is the Conservative Treatment of Infection or Gangrene in Diabetic Patients Worth While? Surgery, **8**: 72, 1940.

[17] McLaughlin, Charles W.: Problems in the Surgical Management of Diabetic Gangrene. Surgery, **13**: 423, 1943.

[18] Joslin, Elliott P.: Proceedings of N. Y. Diabetic Association. Am. J. Digest. Dis., **11**: 7, 1944.

[19] McKittrick, Leland S.: Indications for Amputation in Progressive Arterial Obliteration of the Lower Extremities. Ann. Surg., **102**: 342, 1935.

[20] McKittrick, L., and H. Root: Diabetic Surgery. Philadelphia: Lea and Febiger, 1928.

[21] Smith, Beverly Chew: Amputation Through Lower Third of Leg for Diabetic and Arteriosclerotic Gangrene. Arch. Surg., **27**: 267, 1933.

[22] Maes, Urban: The Surgery of Diabetes as It Concerns Gangrene of the Lower Extremities and Carbuncles. Surg., Gynec. & Obst., **51**: 700, 1930.

[23] Bickel, William H.: Amputations Below the Knee in Occlusive Arterial Diseases. Surg. Clinics of North America, **23**: 982, 1943.

[24] McKittrick, Leland S.: Diabetic Gangrene. Arch. Surg., **40**: 352, 1940.

JUVENILE NASOPHARYNGEAL ANGIOFIBROMA

HAYES MARTIN, M.D., F.A.C.S.
HARRY E. EHRLICH, M.D., F.A.C.S.
AND
JULES C. ABELS, M.D.*
NEW YORK, N. Y.

FROM THE HEAD AND NECK SERVICE, MEMORIAL HOSPITAL

JUVENILE NASOPHARYNGEAL ANGIOFIBROMA, though neither a common nor a malignant neoplasm, nevertheless presents serious problems because of the complications which are invariably associated with its growth and its treatment. This tumor, moreover, is of considerable scientific interest in that it is one of the few neoplasms which exhibit a marked sex predilection—etiologically related in this case, we believe, to a sex-endocrine imbalance. The present report is based on an analysis of 29 cases of juvenile nasopharyngeal fibroma observed on the Head and Neck Service at the Memorial Hospital from 1927 to 1946, inclusive.

DEFINITION

Juvenile nasopharyngeal angiofibroma is a specific, highly vascular, noninfiltrating, essentially benign neoplasm, occurring in the nasopharynx or posterior nasal cavity of pubescent males. Symptomatically the tumor is characterized by nasal obstruction, repeated epistaxis, and by progressive growth throughout the period of adolescence, with a tendency toward spontaneous regression at about the time of sexual maturity.

While the term *juvenile nasopharyngeal angiofibroma* is anatomically specific and descriptive for the tumor, it is somewhat cumbersome, and for the sake of convenience in the remainder of this report, it will often be shortened to *nasopharyngeal fibroma.* Other terms which have been employed elsewhere to designate this neoplasm include *myxofibroma, juvenile basal fibroma, nasopharyngeal fibroma of adolescence, fibroids of the nasopharynx,* and *bleeding fibromas of adolescence.*†

---

* Dr. Jules Abels died June 13, 1947.

† *Historical Note.*—The earliest Greek, Roman, and Arabian medical writers used the term *nasal polyps* (Zool—many footed) to designate all tumors or swellings within the nasal cavities or nasopharynx which caused obstruction to breathing.[1,3] Subsequent medical writers employed the same term for several hundred years. As time passed, more than one variety of nasal polyps were recognized and by the 18th Century such lesions were often differentiated as soft or mucous and hard or fibrous nasal polyps; finally, they were classified into three groups—(1) soft (2) fibrous and (3) malignant. It is somewhat difficult to ascertain just when a specific form of nasopharyngeal fibroma of adolescence was first recognized, but as early as 1847 Chelius[4] stated that fibrous nasal polyps "commonly occur in persons about the time of puberty."

ETIOLOGY

Nasopharyngeal fibroma is not a common tumor. In the Head and Neck Clinic of the Memorial Hospital about 2,000 new cases of neoplasms in the tissues of the head and neck are admitted annually, and of these there are usually one or two cases of nasopharyngeal fibroma. In the literature, most of the case reports number from one to three. A few fairly large series have been published, namely those of New and Figi[21, 9] from the Mayo Clinic — 63 cases, and of Shaheen[6] — 58 cases. Some aspects of the clinical material in these two reports, especially with regard to the ages of the patients and the fairly large proportion of females, suggest to us, for reasons to be considered later, that the cases have not been critically selected and that not all are actually juvenile nasopharyngeal fibromas as defined in the present report.

Age Incidence.—In our series of 29 cases, the age at the onset of symptoms varied from 7 to 19 years with an average age of 14 years. The age on admission averaged about 1½ to 2 years later—that is about 16 years, which is a little younger than Figi's[9] figure of 18½ years at the time of the first examination. In one of our cases, the patient was admitted at the age of 36 years, but had symptoms since the age of 16.

The onset of nasopharyngeal fibroma always occurs during adolescence and is one of its most characteristic clinical features. While many observers,

Considerable space is given in the surgical texts of the early 19th Century to operations for the removal of bulky nasal polyps, especially when the tumors became so large as to produce what was known as a "frog face" deformity. Nevertheless, surgeons of that period, and some even up to the year 1900, appear to have been unaware of any age or sex predilection for these "fibrous nasal polyps." Legouest[17] in 1865 was one of the earliest to call attention to the selectivity of these tumors for males. Gosselin[11] in 1876 noted a tendency toward spontaneous regression after sexual maturity. The first comprehensive study of nasopharyngeal fibroma is to be found in a review on the general subject of nasal polyps in 1878 by Bensch,[2] who collected the scattered case reports and made an excellent morphologic and clinical description of this neoplasm. Chaveau[3] in 1906 suggested the term *juvenile nasopharyngeal fibroma.*

The first attempts at surgical removal of these tumors were made as early as the time of Celsus, by digital manipulations or by tearing off the tumor masses with forceps or snares. Later, injections of various escharotics and the actual cautery were employed.

About the middle of the 19th Century, several operative technics were devised to gain better access to the nasal cavities and the nasopharynx. Langenbeck[16] proposed an anterior approach through the skin of the cheek leaving the bone attached to the osteoplastic flap and temporarily resecting the maxilla. After removal of the tumor, the flap with the attached bone was replaced and the wound was sutured. To obtain access to the nasopharynx Rouge[25] reflected the nose upward and resected part of the septum by an incision in the upper gingivo-buccal gutter. Ollier[22] made a V-shaped incision with its base opposite the nasal ala, reflecting the whole nose downward. All of these operations were rather bloody and the mortality was high. Bilateral ligation of the external carotid arteries and tracheostomy were mentioned as preliminary surgical measures as early as the 1880's.

Most of the reports in the literature consist of three or less cases and the clinical data are often meagre and uncertain. In recent years several larger series have been published which we shall discuss later in greater detail.

as far back as Chelius[4] in 1847, have believed that this neoplasm occurs mainly in children, hence the eponym "juvenile," nevertheless such observers as New and Figi[21, 9] and Shaheen[26] have included cases with a supposed onset long after sexual maturity. Some of these apparent variations may arise by recording the age of the patient on admission rather than the age at the onset of symptoms. Shaheen[26] is the only recent observer who has reported the occurrence of this tumor during infancy and old age (2 and 4 years, 67 and 70 years, respectively). Since Shaheen appears to be unaware that his reports of cases occurring at the age extremes are at variance with those of most other observers and since he offers no histologic or other corroborant data, we feel that these particular cases are not all acceptable as genuine instances of juvenile nasopharyngeal fibroma.

The duration of the tumor before it produces symptoms cannot of course be determined. From the observed rate of growth following the first examination in untreated patients, however, it is probable that a silent period of 2 to 3 years may elapse, bringing the onset of the tumor to between 11 and 12 years. We have not been able to find any comment on this probability in other reports.

Sex Relationship.—In the course of the present study, certain sex-endocrine factors of probable etiologic significance became apparent. These were:

1. The disorder was limited to young males.*

2. These patients in most instances gave the clinical impression of undersexual development, both physically and emotionally.

3. In spite of roentgen-ray therapy the tumor significantly regressed only after secondary sex characteristics were developed fully; there was one exception in the 29 cases presented.

4. In two instances in which puberty was hastened by the administration of androgens, roentgen-radiation appeared to induce a more ready regression of the tumor.

* All of our 29 patients with nasopharyngeal fibroma were males and we are of the opinion that this neoplasm never occurs in females. Beginning with Legouest[17] in 1865, many subsequent authors have recognized that the incidence of this tumor was considerably greater in males; but we are the first, so far as we know, to advance the proposition that juvenile nasopharyngeal fibroma is a completely sex-bound neoplasm. We realize that this point of view may not be generally accepted without question at this time, for all previous authors who have reported large series have given a definite percentage of female incidence (Figi and New, 7 per cent; Shaheen, 8 per cent). At the Memorial Hospital, up until about 10 years ago, we also believed that juvenile nasopharyngeal fibroma occurred occasionally in females, but since that time we have subjected all cases so diagnosed in female children to careful scrutiny. These investigations, based not only on biopsy but also on the subsequent clinical course, have failed to support the diagnosis of nasopharyngeal fibroma in a single female patient in our clinic. In three suspected cases in females the lesions on biopsy proved to be simple choanal polyps. In two other cases the eventual diagnosis was tuberculosis of a retropharyngeal lymph node and chondroma arising in a superior nasal turbinate, respectively. In none of these tentatively diagnosed cases of juvenile nasopharyngeal fibroma in females were found the characteristic symptoms and clinical course—progressive nasal obstruction, recurrent epistaxis, and spontaneous regression at sexual maturity.

These observations strongly suggest that juvenile nasopharyngeal fibromas may result from a deficiency of androgen activity or, perhaps, from an overproduction of estrogens. Unfortunately, little or no information is available concerning estrogen production by pubescent males from which conclusions might be drawn.

There is, however, considerable clinical and experimental evidence that vascular tissue can be influenced by certain of the sex hormones, but these effects are not always uniform. In cutaneous areas characterized by a large venous bed, the capillaries of *castrated* males are found widely dilated and this dilatation can be reversed by the administration of testosterone propionate.[7] Likewise, the excitability of cutaneous blood vessels has been found to be more extensive in castrated men and this excitability to graded mechanical stimuli could be increased by estradiol and decreased by testosterone proprionate.[24]

The observations of Soskin and Bernheimer[27] that relief of atrophic rhinitis was obtained by estrogen administration stimulated several studies concerning the relation of sex hormones to mucous membrane hyperemia. Of these, the work of Reynolds and his co-workers[23] bears most on the present clinical study. These investigators demonstrated that hyperemia of the mucous membranes was a function of blood estrogen content and could be induced by further estrogen administration. Furthermore, the mechanism of the hormone action was discovered to be due probably to the local production of acetylcholine. The clinical and experimental value of these observations have been demonstrated in the treatment of peripheral vascular disease. For example, gangrene induced by ergot drugs can be prevented by estrogen administration.[18]

During the course of further studies it is planned to give much more consideration to the sex linked character of this tumor. Particular attention will be given to:

1. The determination of "developmental age" of these patients by roentgen-ray examination of ossification centers.
2. The excretion of total 17-ketosteroids; for this a considerable amount of work first must be done to establish the normal range for the 17-ketosteroid excretion of the pubescent male.
3. The effects of androgens alone in massive doses on the tumor before roentgen-radiation or surgery is applied.
4. The effects of estrogens on the appearance of the tumor.

Histogenesis.—Histologic study in our cases reveals that in the tumors of younger subjects the angiomatous elements predominate and occasionally a microscopic picture of fully developed cavernous angioma is noted. It is probable, therefore, that in the beginning these growths are principally angiomatous rather than fibromatous. If estrogenic stimulation is a factor in the etiology of these tumors, their histogenesis can be reasonably explained as an overgrowth of vascular tissue in the nasopharynx, a result of an abnormal

stimulus to the local circulation. The fibromatous elements, at first being only supporting stroma of the tumor, develop as a structural component and become predominant as the estrogenic effect is lessened. An identical phenomenon is the gradual replacement of the angiomatous elements by fibrous tissue as seen in ordinary hemangiomas following spontaneous or therapeutically induced sclerosis of the blood vessels.

When the patient approaches sexual maturity, as will be later described, all tumors tend to become less vascular and often completely fibrous, which suggests that as the abnormal stimulus disappears the proliferation of blood vessels also ceases. This hypothesis, if correct, is sufficient to explain the histogenesis of this tumor.

Other genetic theories have been advanced which presuppose that the growth is of fibroblastic origin. The most prevalent and hitherto accepted explanation of the origin of nasopharyngeal fibroma, first advocated by Verneuil[29] and supported by Bensch,[2] Ewing,[8] and others, is that the growth is derived from embryologic fibrocartilage during development of the skull. The embryonal occipital plate, a cartilaginous structure, gives rise to the basilar portion of the occipital bone, body of the sphenoid bone, medial pterygoid process, and bones in the region of the foramen lacerum and pterygopalatine space. Until early adult life the basilar portion of the occipital bone is joined to the body of the sphenoid bone by the remaining portion of embryonal cartilage. This cartilaginous plate becomes ossified by the 25th year. According to these authorities, perichondrium (fibrous connective tissue) covers the cartilaginous plate and from this or other perichondrium of the postnasal space juvenile nasopharyngeal fibroma is supposed to develop. This hypothesis is attractive in that it accounts for the phenomenon of spontaneous regression of the tumor about or after the 25th year, and also, for its various anatomic sites of origin. The latter theory, however, does not explain the sexual selectivity of this tumor nor does it take into consideration the presence of angiomatous elements which are integral and, perhaps, the more significant morphologic components of nasopharyngeal fibroma.

In 1943 a case of chondrosarcoma of the nasopharynx occurring in an adolescent boy was reported by Wirth.[28] Although this tumor finally metastasized to the lungs as chondrosarcoma, the initial biopsy was compatible with a diagnosis of juvenile nasopharyngeal angiofibroma. The presence of cartilage even in the first biopsy specimen, the lack of response of the growth to large doses of external and interstitial radiation, and subsequent biopsy reports (vascular embryonal chondroma; chondrosarcoma) mitigate against this growth being a genuine nasopharyngeal fibroma in the beginning, as Wirth admits. If it could be proved, however, that chondrosarcomatous transformation can occur in a pre-existing nasopharyngeal fibroma, it would favor the prechordal plate genetic theory of Verneuil.

Other Causative Factors.—Further analysis of our data reveals no evidence of any systemic disturbances. The majority of patients were investi-

gated for syphilis, tuberculosis, and other infections and were found to be free from these diseases. Allergy or trauma did not appear to be etiologic factors. Congenital anomalies, mal-developments and other associated tumors were not encountered and in no case were either neurofibromatosis or hemangiomatous tendencies present. The boys were not retarded, either physically or mentally, and except for evidence of underdevelopment of secondary sex characteristics in over one-half of the cases, they appeared normal in all other respects.

No tendency toward racial or familial predilection was noted in the present series or in any previously reported cases.

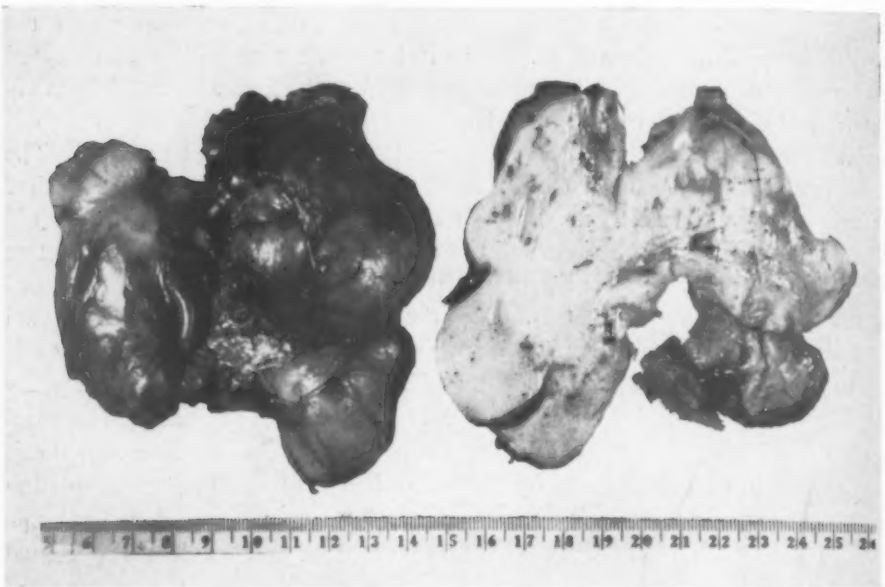


FIG. 1.—Surgical Specimen of Nasopharyngeal Fibroma. This bulky, lobulated, cartilaginous-like mass was removed from the nasopharynx and posterior nasal cavity of an 11-year-old boy by a radical Weber-Ferguson type of resection of the maxilla. Note that the tumor is dumb-bell shaped; one portion projected into the posterior nasal cavity and the other beneath the soft parts of the cheek in the infrazygomatic and temporal areas.

PATHOLOGY

Gross Pathologic Anatomy.—Nasopharyngeal fibroma is an unencapsulated, fungating, vascular tumor. The surface of the growth, if not traumatized by operative intervention or packing to control hemorrhage, is covered by intact mucous membrane, highly injected and deep red in color in younger subjects and pale pink in older patients or those in whom the vascularity has been reduced by radiation or sex hormone therapy. If there has been hemorrhage with the attendant trauma of packing, ulceration and necrosis occur and the surface of the tumor may become granular.

In our cases, with one execution, the growth ranged from 2 to 5 cm. in

greatest diameter and the average size was 3 cm. One tumor was so large (10 cm.) as to be out of proportion to the standard variation in this series.

In the specimens available for gross morphologic study, unusual variations and combinations were noted. Although the shapes of the tumors differed widely, the commonest types were ovoid or club-shaped. The surface was either smooth or definitely lobulated (Fig. 1). Some of the tumors were rubbery or cartilaginoid in consistency while others were soft, edematous and occasionally friable. On section the color of the neoplasms ranged from pinkish white to grayish yellow to reddish brown, and translucent tissue was seen as often as homogeneous tissue. All growths were solid with no areas of

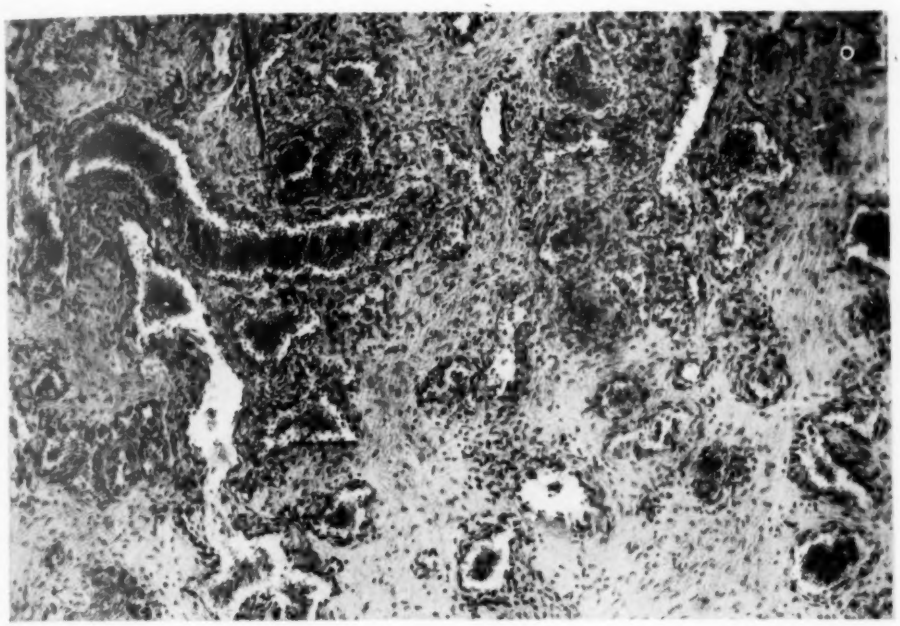


FIG. 2.—Vascular Phase of Nasopharyngeal Fibroma. In this microphotograph of an extremely vascular nasopharyngeal fibroma in a 10-year-old boy, the microscopic appearance is that of a fully developed cavernous angioma in a fibrous stroma. Some of the blood vessels have become sinusoid. This tumor had no previous sex hormone or radiation therapy.

cystic degeneration. Older tumors or those which had been subjected to radiation therapy were usually densely fibrous and pale; younger and untreated tumors were soft and deeply vascular.

Histopathology.—Nasopharyngeal fibroma is composed essentially of connective tissue and blood vessels. In microscopic appearance it may vary from that of a fully developed cavernous angioma in a fibrous stroma (Fig. 2) to that of a densely cellular or occasionally myxomatous fibroma (Fig. 3). The usual histologic pattern consists of connective tissue stroma containing numerous spindle-shaped immature fibroblasts and thin-walled blood vessels in

varying proportions. In fact, spindle- and star-shaped fibroblasts may be so numerous as to suggest fibrosarcoma or angiosarcoma, with which juvenile nasopharyngeal angiofibroma is often confused. In younger subjects and in untreated tumors, the angiomatous elements are in abundance, the vessels becoming large, irregular and even sinusoid; in older tumors or in those which have been subjected to radiation therapy or intensive treatment with androgens, the vascular components are less prominent or they may disappear altogether and fibrous elements predominate. Foci of lymphocytes and plasma cells may be present, especially in those tumors which are ulcerated or traumatized. Myxomatous changes in varying proportions are frequently found,

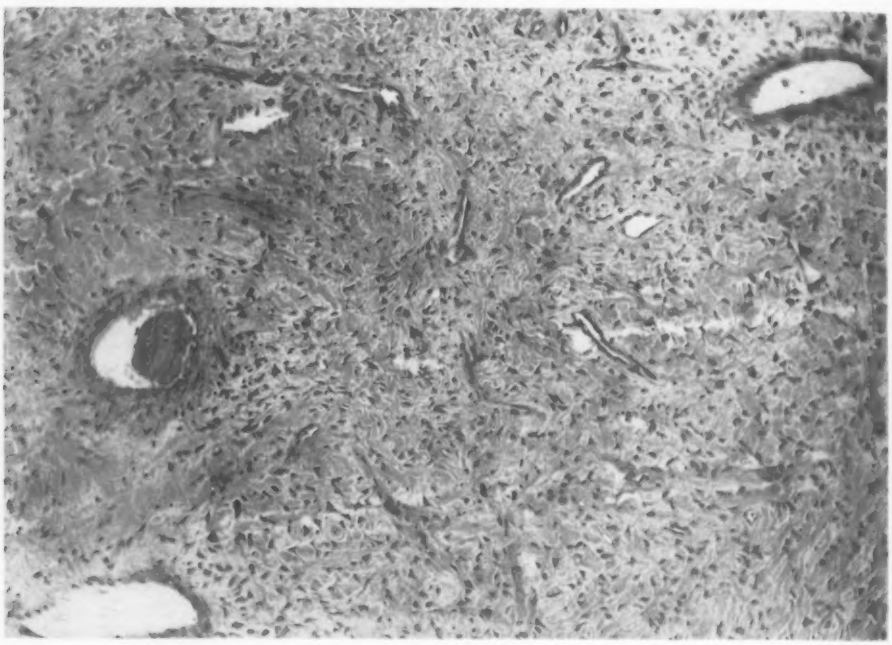


FIG. 3.—Avascular Phase of Nasopharyngeal Fibroma. The microscopic appearance of a relatively avascular nasopharyngeal fibroma consists of sparsely scattered blood vessels in a densely cellular stroma. This histologic pattern of replacement of angiomatous elements by connective tissue (involution) can frequently be brought about by androgenic therapy and irradiation.

as are areas of necrosis. Hyalinized thrombi are occasionally seen, especially in older or treated tumors, together with areas of hyalinized stroma.

The tumor possesses no true capsule. A pseudo-capsule which actually consists of pharyngeal or nasal mucosa, occasionally stretched and atrophic, is noted in non-ulcerated specimens (Fig. 4).

Although malignant transformation has been reported by others (Shaheen,[26] Jackson,[15] Dabney[5]) the presented evidence is inadequate and unconvincing, in our opinion. No tumor in the present series underwent malignant

transformation, anatomically or clinically, with the exception of a single case in which one of numerous recurrences revealed unusual cellularity and localized areas of malignant transformation; subsequent recurrences, however, during a period of three years were reported as benign and there have never been any clinical manifestations of malignant behavior. This case will be referred to again under prognosis.

SYMPTOMS, MORBID ANATOMY AND CLINICAL COURSE

The first symptom of nasopharyngeal fibroma is probably always *nasal obstruction* which, if only moderate in degree, may for a time pass unnoticed.

FIG. 4.—Surface Appearance of Nasopharyngeal Fibroma. The neoplasm has no true capsule. In this microphotograph intact mucosa may be seen stretched over the tumor, forming a pseudocapsule.

While in a few of our cases the patients at first gave epistaxis as the initial symptom, closer questioning almost always elicited a preceding history of nasal obstruction to which little attention had been paid. In order to produce nasal obstruction, we estimate that a tumor in the nasopharynx must reach the size of about 2.5 cm. in diameter, although in the choana a smaller mass could undoubtedly produce this symptom. At any rate there is probably a silent period of at least several months before a growing nasopharyngeal fibroma becomes large enough to cause some obstruction to breathing.

The second symptom of nasopharyngeal fibroma and the one which most often causes the patient to seek medical advice is *recurrent epistaxis*. Hemor-

rhage probably occurs either as a result of trauma to the tumor, incident to sneezing or to forcibly blowing the nose, or from pressure necrosis of the expanding growth as it meets with the resistance of the confining bony walls. Once initiated, hemorrhages occur at increasingly frequent intervals, especially when their control necessitates tamponage or nasal packing. These manipulations, when repeated, almost always result in sepsis which may extend to the paranasal sinuses, the middle ear, and even the mastoid. With recurrent hemorrhages, the patient becomes anemic and since proper nutrition is interfered with by local manipulations, usually loses considerable weight.

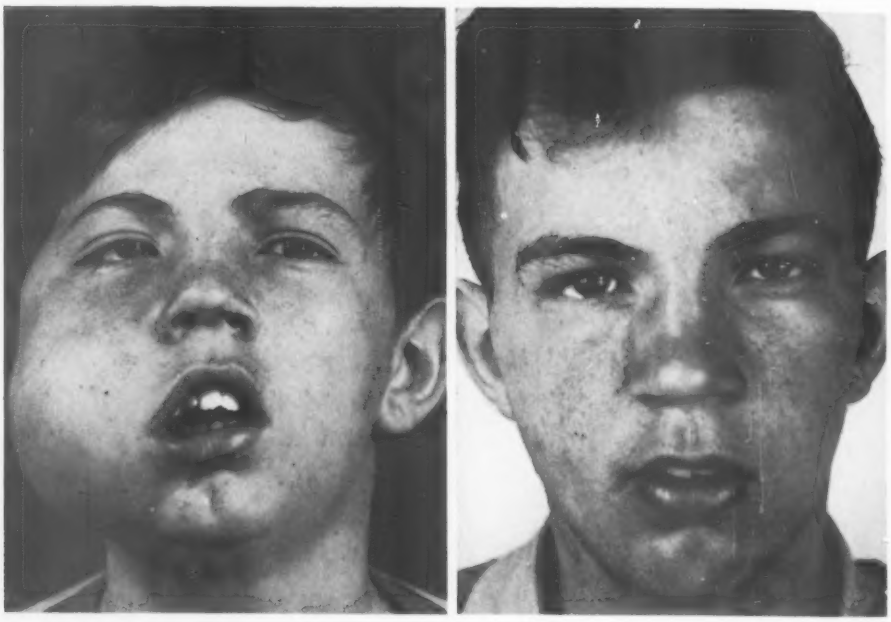


FIG. 5.—Facial Deformity in Nasopharyngeal Fibroma. (a) Marked facial deformity may be produced by a bulky nasopharyngeal fibroma as the mass grows outward compressing the antrum and pushing the soft parts of the cheek ahead of it. Removal of this tumor necessitated resection of the anterior wall of the maxilla and corresponding alveolus after reflecting a cheek flap. (b) Postoperative photograph.

If hemorrhage is not an early or frequent symptom and the nasal obstruction is ignored, the tumor may attain sufficient size to cause "frog face" deformity so often mentioned in the surgical literature of the 19th Century.* Such a degree of facial deformity, consisting of prominence of the cheeks and

* "Frog-face" deformity is a clinical manifestation associated with lesions other than juvenile nasopharyngeal angiofibroma. Benign and malignant neoplasms of the nasal cavity, nasopharynx, and maxilla not infrequently occur in children, such as ossifying fibroma, sarcoma of the soft parts, lymphomatous tumors, and central myxoma. As the growth expands and advances, the floor of the orbit is elevated (unilateral or bilateral), producing "frog-face" deformity.

nose, obliteration of the naso-labial groove, and prominence of the eyes, was present in about one-quarter of our cases on admission (Fig. 5). The character of the deformity results not only from the expansion of a growing bulky tumor but sometimes from the particular direction that a growth has taken from its point of origin.

Most of the tumors appear to have origin in the vault of the nasopharynx or at least mainly in the vault and the immediately adjacent lateral or posterior nasopharyngeal walls. The base of attachment of the growth is usually rather broad (2–3 cm. or more) so that the exact point of origin and extent of attachment is difficult to delineate either by clinical examination or operative exposure.

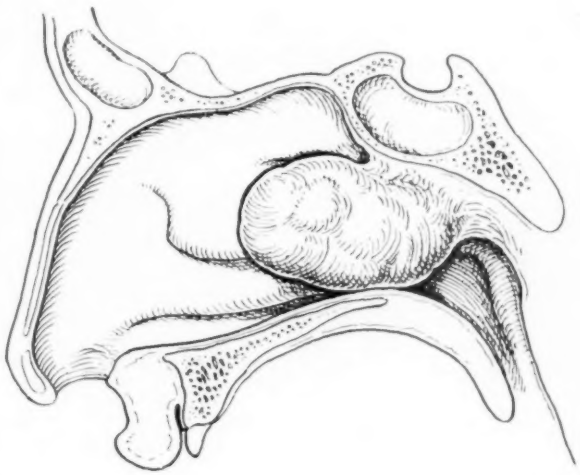


FIG. 6.—Anterior Growth of Nasopharyngeal Fibroma. Diagram of nasopharynx (sagittal view) showing a common location for nasopharyngeal fibroma. The tumor arises from one of the walls of the nasopharyngeal cavity or posterior nasal space and grows downward and forward blocking one or both choanae.

As the tumor enlarges and expands it may grow forward into the nasal cavity (Fig. 6), often presenting at one anterior naris as an edematous partly necrotic mass; in other cases the growth extends backward to protrude below the free edge of the soft palate; some proceed laterally, perforating by pressure necrosis the maxilla to enter the antrum and even appearing in the subcutaneous tissues of the cheek (Fig. 7). A large growth occasionally produces pressure on the floor of the orbit and causes elevation of the globe with resultant unilateral exophthalmos. In any case, the main mass is always in the nasopharynx.

The osseous walls of the nasopharynx are formed by the body of the sphenoid bone, the basilar portion of the occipital bone, the medial pterygoid

plate, and the cervical portion of the vertebral column. The union of these bones, the fascial and tendonous structures which are attached to them, together with variously sized recesses produced by numerous mucosal folds and the cartilages of the eustachian tube (torus tubarius, fossa of Rosenmüller, etc.), contribute to the unusual irregularity of the walls of the nasopharyngeal cavity. This irregularity makes complete surgical removal of a densely adherent broadly attached tumor, like nasopharyngeal fibroma, difficult if not impossible. The peculiarities of the surgical anatomy of nasopharyngeal fibroma will again be referred to in the section on treatment.

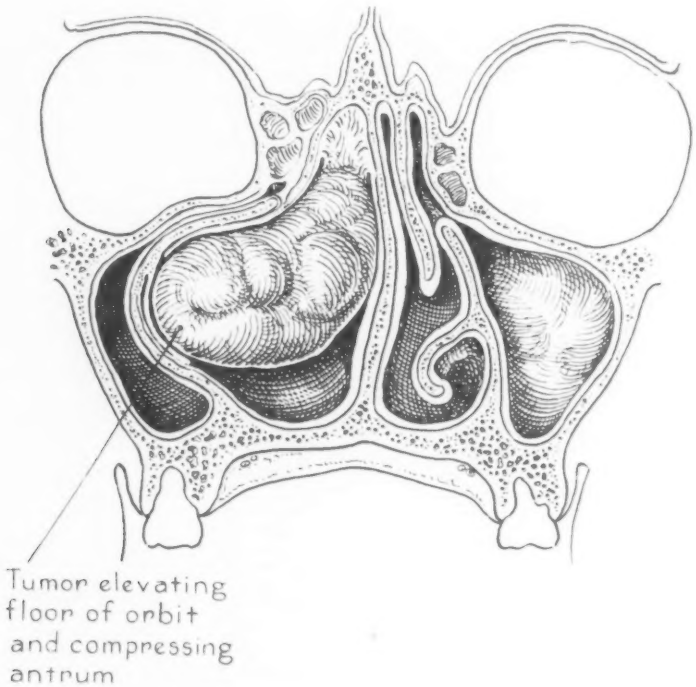


FIG. 7.—Lateral Growth of Nasopharyngeal Fibroma. Diagram of nasopharynx (coronal section) showing direction of growth frequently taken by nasopharyngeal fibroma. After plugging the posterior nasal cavity, the tumor compresses the antrum, expands laterally into the maxilla, and may eventually reach the soft tissues of the cheek.

As an expanding tumor closes off the nasopharynx, voice changes may occur (*rhinolalia clausa*) and, also, a loss of sense of smell. In our series, there is no instance of erosion of the base of the skull, although it has been described by Goldsmith.[10]

The usual history in patients with juvenile nasopharyngeal fibroma applying to Memorial Hospital followed a rather definite pattern: an acute onset of recurrent epistaxis in boys from 12 to 15 years of age, usually preceded by

progressive nasal obstruction to which little or no attention had been paid. With each incidence of epistaxis, the nose had been packed and the hemorrhage temporarily arrested, only to recur with increasing frequency and severity. In most of the protracted cases there was marked local sepsis and sometimes partial necrosis of the tumor.

In about one-half of the cases in our series, previous operative attempts to remove the tumor had been made, frequently through the mouth and sometimes by splitting the soft palate. Usually two or three and in one case five operative efforts had been made. In almost every instance where surgical removal had been undertaken, the operator admitted that he had abandoned the attempt to completely remove the growth because of ensuing hemorrhage. In about 50 per cent of these failures, the diagnosis of "sarcoma" was made, the case was given up as hopeless, and the patient was referred to Memorial Hospital for palliative roentgen-ray therapy. In some of the cases, prior to referral to us, snare removal of a "nasal polyp" had been tried, or the bleeding point had been cauterized, or, in five cases, tonsillectomy and adenoidectomy had been performed in the belief that the tonsillar hypertrophy was the cause of the symptoms. All of these efforts had been followed by an increase in nasal bleeding.

In advanced and complicated cases such as described above, especially after incomplete surgical removal, there is usually an ill-defined, bulky, infected, partly necrotic, vascular tumor filling the nasopharynx and extending into one or both nasal cavities. The picture is sometimes one of subacute sepsis, pansinusitis, otitis media, mastoiditis, anemia, and malnutrition which, if unrelieved, results fatally despite the fact that the neoplasm is essentially benign and self-limited in growth.

In contrast to the tragic picture of the advanced and neglected cases, many others, properly managed, follow a relatively benign course provided that the symptoms of an expanding growth and hemorrhage can be kept under control. It is probable that some cases of nasopharyngeal fibroma of moderate size occur and regress spontaneously without ever being discovered or producing any marked symptoms. We have under observation at Memorial Hospital a case in which a nasopharyngeal tumor, undoubtedly a nasopharyngeal fibroma, was discovered by us on routine physical examination in a 15-year-old boy. The growth was not large enough to produce nasal obstruction and there had been no epistaxis. No treatment has been given. The tumor has been observed for two and one-half years and has remained about stationary in size. It is probable that such asymptomatic nasopharyngeal fibromas may often occur and regress when sexual maturity is reached.

DIAGNOSIS

The histories of the 29 cases in the present series reveal that the correct diagnosis was seldom made by the physician first consulted. In many instances a clinical diagnosis of malignant tumor was made and without further inves-

tigation radiation therapy was advised; in other cases, after an unsuccessful attempt to remove the growth or the adenoids had been made, resulting in profuse hemorrhage, the patient was referred to us for treatment of "sarcoma." Such defeatist attitudes were the rule if the boy presented appreciable facial deformity or proptosis. Only occasionally, when a biopsy had been performed, had an accurate diagnosis of juvenile nasopharyngeal fibroma been made.

Although the tumor is admittedly rare and on the basis of incidence will not be recognized by the casual examiner, nevertheless the unique anatomic and clinical setting of this neoplasm should plainly suggest the possibility of nasopharyngeal fibroma. The clinical syndrome of rapidly progressive nasal obstruction, recurrent severe nosebleeds in *pubescent and adolescent males*, plus the presence of a discrete, ovoid or club-shaped, smooth, vascular and usually bulky nasopharyngeal mass, which has grown forward to block one or both choanae, justifiably warrants a clinical diagnosis of nasopharyngeal fibroma. It should be emphasized, however, that only by mirror examination of the nasopharynx through the open mouth, occasionally supplemented by direct rhinopharyngoscopic examination through the anterior nares, can the tumor be visualized and its essential character appreciated.

Stereoscopic radiographic examination of the base of the skull, paranasal sinuses, and nasopharynx is not only of value in determining the exact location and extent of the tumor, but is especially important in the differential diagnosis where a bulky growth in the head has brought about considerable deformity of the face or unilateral exophthalmos.

Differential Diagnosis. There are several extra-nasopharyngeal lesions which produce either nasal obstruction, epistaxis, unilateral exophthalmos, deafness, or facial asymmetry, singly or in combinations. Asymptomatic tumefactions may also be encountered in the nasopharyngeal or posterior nasal cavity which are occasionally mistaken for juvenile nasopharyngeal fibroma.

Choanal polyps can be distinguished from nasopharyngeal fibroma even when they occur in boys. These growths are frequently multiple, bilateral, pedunculated and tend to appear in individuals suffering from an allergy. They rarely exceed 1.5 to 2 cm. in diameter and resemble in every way the common and well-known pale, edematous polyps found in the anterior nasal cavities. Microscopic examination of the tissue conclusively establishes the diagnosis.

Pharyngeal tonsil (adenoids) is a common cause of nasal obstruction and nasal speech in juveniles. Hyperplastic lymphoid or granulation tissue in the nasopharynx may even be responsible for recurrent nosebleeds, although rarely profuse. There should be no difficulty in differentiating this condition from nasopharyngeal fibroma inasmuch as aggregates of lymphadenoid tissue, generally on the posterior nasopharyngeal wall, present a typical appearance consisting of irregular, conglomerate, soft, grayish-red masses which are distributed along the mucosa.

Benign minor salivary gland tumors, carcinoma, and malignant lymphomas, though uncommon in the head and neck of children and adolescents,

occur with much greater frequency in the nasopharynx and posterior nasal cavity than nasopharyngeal fibroma and, although they may produce symptoms similar to those of nasopharyngeal fibroma, in no way resemble it on clinical examination.

Chordoma of the upper cervical vertebrae, malignant tumors of the nasal accessory sinuses, and primary osseous neoplasms of the maxilla may produce nasal obstruction, epistaxis, and facial asymmetry, but by careful physical and roentgenographic examination these lesions can be tentatively diagnosed and differentiated from nasopharyngeal fibroma with little difficulty. We have seen chondroma in the posterior nasal cavity, tuberculosis of a retropharyngeal lymph node, and olfactory aesthesioneuroblastoma in the postnasal space, which clinically resembled nasopharyngeal fibroma in every way.

Biopsy.—An effort should always be made to establish the diagnosis by biopsy. It is prudent to hospitalize the patient for this procedure so that, if necessary, satisfactory anesthesia and facilities for control of profuse hemorrhage and for blood transfusions can be available. Deferment of biopsy is often wise if the patient is either bleeding actively or still recuperating from severe epistaxis or if there is appreciable infection in the nasopharynx, paranasal sinuses, or middle ear.

A straight biopsy forceps inserted directly backward through the nasal cavity or a curved forceps inserted through the open mouth may be used to remove a specimen from a nasopharyngeal tumor. The manipulations in either instance should be guided visually by a mirror or digitally by a finger.

Although biopsy is highly desirable and usually essential in the diagnosis of neoplasms, it must be conceded that in some cases of nasopharyngeal fibroma, despite the best of intentions, a positive histologic report cannot be obtained. In some cases, a recurrent growth, after several operative attempts, is largely necrotic and the local condition in the nasopharynx is one of advanced sepsis. Repeated biopsies in such instances may show nothing more than necrosis or granulation tissue. If the case is clinically typical in all other respects (age, sex, characteristic sequence of symptoms, anatomic location, gross appearance of the tumor), the diagnosis of nasopharyngeal fibroma may fairly be made even though not confirmed by a positive biopsy.

In our clinic where primary histologic confirmation is insisted upon, we nevertheless felt justified in making a diagnosis of nasopharyngeal fibroma in five cases in which a positive microscopic report on the biopsy specimen could not be obtained, despite repeated attempts at biopsy in three of these. The subsequent clinical course of these patients confirmed our initial belief that we were unquestionably dealing with nasopharyngeal fibroma.

TREATMENT

A basic consideration in the treatment of nasopharyngeal fibroma is the fact that this tumor is anatomically and clinically benign, that with few exceptions it will begin to regress spontaneously at about the time of sexual maturity, and from then on will cause no further trouble. The real hazards consist

of the complications—hemorrhage, sepsis, facial deformity, and last but not least the effects of injudicious and over-aggressive attempts at complete eradication of the growth by surgery or radiation therapy. Provided that the aforementioned complications can be kept under reasonable control during the period of adolescence by moderate irradiation or limited surgery, and sex hormone therapy, spontaneous regression almost uniformly occurs at the time of sexual maturity.

The types of deforming surgical procedures particularly to be avoided in nasopharyngeal fibroma are approaches through the skin of the middle of the cheek (Langenbeck[16]), temporary detachment of bone (Ollier[22]) or exposure of the nasopharynx by splitting the soft palate. We shall discuss the more restrained and judicious forms of surgical procedure later in this report.

By over-aggressive and injudicious radiation therapy we mean any dose of interstitial radium (or radon) which is likely to produce radionecrosis in the tumor or adjacent palate, skin of the face or bones of the skull, or the application of roentgen-radiation in such dosages and through ports so located and of such size as to arrest or markedly retard the growth of the maxillae and other facial bones.

Once a diagnosis of nasopharyngeal fibroma is made, a systematic plan of management should be instituted, consisting of a combination of at least two and sometimes more methods of treatment. First, if epistaxis has been marked, both external carotid arteries should be ligated and measures taken to improve the local hygiene, dispensing, if possible, with any nasal packing or tamponage which inevitably incites a vicious cycle of further hemorrhage, repeated tamponage, and sepsis. Next, the administration of androgens adequate to induce the development of secondary sex characteristics without undue emotional imbalance should be instituted. The amounts of testosterone propionate and/or methyl testosterone required probably are very different for each individual and may be a function of his chronologic and "developmental" ages. At the same time radiation therapy (radium or roentgen-ray or both) should be given. Lastly, if the tumor is so large as to produce complete blockage of the nasal cavity with an edematous, partly necrotic mass or with symptoms of pansinusitis, then some form of partial surgical removal should be considered. The rationale and technic of these several therapeutic programs will next be discussed separately and in detail.

Ligation of the External Carotid Arteries.—This is one of the most direct and useful procedures in controlling arterial hemorrhages from any portion of the mouth, middle or upper pharynx, and nasal cavities. It is an entirely safe and harmless procedure to ligate permanently both external carotid arteries at one operation. Ligation of these blood vessels will not always completely control hemorrhage from the nasal cavities and nasopharynx, but it will markedly reduce it.

While a portion of the arterial supply of the nasal cavities is derived from the internal carotid artery by way of the ophthalmic artery and its anterior and posterior ethmoidal branches, this source can hardly cause any major difficulty.

A much more troublesome source of hemorrhage from the nasopharynx and nasal cavities is venous—from the pterygoid plexus which communicates not only with the anterior facial vein but also with the veins of the cranial cavity by way of the superior ophthalmic vein. In any case, as compared with arterial hemorrhage, venous bleeding tends to be less profuse and the relatively low venous pressure may more easily be controlled by temporary light tamponage.

Sex Hormone Therapy.—This form of therapy can be started immediately in all cases without interfering with any other treatment measure. A clinical evaluation of the status of sexual development should be made by frequent examinations to determine any change possibly brought about by androgenic therapy. The state of the ossification centers, the level of urinary excretion of 17-ketosteroids, careful evaluation of secondary sex characteristics and emotional changes should be made at frequent intervals.

We had hoped at first to be able to control nasopharyngeal fibroma by endocrine therapy alone. This has not proved possible in the few instances in which its limited use has been employed. We have noted in these instances a definite and gradual elimination of the hemorrhagic tendency of the tumors associated with an acceleration of sexual maturation. In particular this association was observed in the case of an 18-year-old male who was treated for several months with limited benefit by radiation therapy. The intramuscular administration of 25 mg. of testosterone proprionate four times each week effected a marked change of his secondary sex characteristics and a rapid disappearance of the tumor in the space of one month.

Roentgen-ray Therapy.—Practically all hemangiomatous lesions are at least moderately radiosensitive. Fibrous tissue and fibromas in general are not radiosensitive. In our opinion radiation therapy is useful in nasopharyngeal fibroma mainly as a measure to reduce the angiomatous component of the tumor, thereby assisting in the control of the hemorrhage and to some extent in the arrest of its growth. We think it doubtful that radiation therapy in justifiable dosage can have much direct effect on the fibromatous elements of this neoplasm.

Radiation therapy is immediately indicated in bleeding nasopharyngeal fibroma but is of less value as initial treatment in bulky (6–8 cm.) edematous tumors in which the clinical picture is mainly one of facial deformity and nasal obstruction from an expanding tumor. Roentgen-ray therapy can be instituted promptly, preferably through the open mouth and hard palate in posteriorly placed growths. Additional roentgen therapy may also be given externally through the maxillae, but here some thought should be given to the possibility of permanent damage to the development of the facial bones. We have never been able to determine exactly what dose of roentgen therapy can be given safely over the germinal centers of growing bones. In general, we suggest in nasopharyngeal fibroma not more than 1000–1500 r in divided doses through circular portals 5 cm. in diameter over each maxilla (200–250 K.V., 50 cm. T.S.D., 1–1½ mm. copper filtration). Even this dose may result in some

flattening of the cheeks as the boy reaches full stature. The dose through the hard palate may be greater (2000–2500 r) with less danger of late deformity, employing peroral circular or oval portals 3–4 cm. in diameter. By cross-firing the tumor through these three portals, a significant roentgen-ray dose can be delivered into the tumor site.

Radium Therapy.—If the growth is bulky (5 cm. or more in diameter), no significant radiation dose which may be considered safe can be distributed throughout the mass by interstitial radium or radon. If the tumor is extremely vascular, the insertion of a trocar for implantation of seeds or the introduction of a radium needle is attended by profuse hemorrhage, necessitating immediate and firm tamponage. Under such conditions, the procedure appears to cause as much harm as good even though the radiating sources remain in place.

Radium therapy in the form of gold radon implants is one of the most useful methods for control of nasopharyngeal fibroma provided the tumor is less than 5 cm. in diameter and the dosage is fractionated and combined with other forms of treatment such as supplemental roentgen-radiation to the cheeks and palate and hormone therapy. Seeds in doses of 5–6 mcs. (unit strength 1–1.5 mcs.) can be inserted either through the nasal cavity and/or the soft palate with the aid of the exploring finger passed behind the soft palate into the nasopharynx. The dose may be repeated once or twice at intervals of about a month. Delevan[6] and Figi[9] have recommended that the main reliance be placed on this method of treatment. Radon seeds are also useful for the treatment of residual tumor or re-growth after surgical removal of large neoplasms which were too bulky for control by radiation therapy in the beginning.

Surgical Excision.—It is not possible to obtain even a moderately wide surgical exposure of the nasopharynx except by operative procedures which are not only immediately hazardous but also permanently deforming and disabling. Whether the surgical approach is made through the maxilla anteriorly, the alveolar process, or the palate, the permissible size of the opening through the bone is limited to 3–4 cm., and the destination of the approach, that is the cavity of the nasopharynx, is at a depth of 9–10 cm. from the surface. Deliberate and continued surgical dissection at such a depth in a highly vascular zone and through such a narrow aperature is simply not possible. For these technical reasons, and the almost inevitable postoperative recurrence, surgical removal of nasopharyngeal fibroma, except when absolutely necessary, has never been regarded enthusiastically by experienced observers. Nevertheless, in the larger tumors with marked facial deformity and progressive destruction of the maxilla or other adjacent bony structures by pressure necrosis, the bulk of the tumor should be removed or at least reduced by some expedient other than radiation therapy. To this end several methods have been employed by others, such as avulsion by wire snare through the nasal cavity or mouth, destruction of the tumor *in situ* by endothermy, and by the direct trans-maxillary surgical approach.

We have used several methods for the surgical removal of these and other varieties of tumors in the naso-pharynx, the selection depending mainly on the position and surgical anatomy of the growth. In some cases where the nasopharyngeal fibroma was not too large, we have employed the wire snare or simple avulsion by digital manipulations through the anterior nares and mouth. Such simple maneuvers have at least the advantage of being nondeforming. In others, we have approached the nasal cavity and nasopharynx through an incision in the upper gingivobuccal gutter (Rouge), but have found that unless a considerable portion of the nasal bones and nasal septum are rongeured away this approach has little advantage over simple dilation of the anterior nares. In some cases where the tumor extended directly forward through the antrum and anterior wall of the maxilla, we have developed and reflected a cheek flap (Weber-Ferguson incision), exposing the tumor directly from the front. One of the most useful, least deforming, and least disabling approaches to the nasal cavity and nasopharynx is through the alveolar process anteriorly, just to one or the other side of the midline. If teeth are present, about 4 or 5 are extracted, and the entire thickness of the alevolus removed exposing both the antrum and nasal cavity on that side. The party wall between the nasal cavity and the antrum is then rongeured away and a fairly wide exposure of the nasopharynx obtained with adequate accessibility to the ethmoid and sphenoid sinuses. The latter operation is followed by surprisingly little disability. The opening in the alveolus shrinks down to about 2 cm. in diameter and a dentist can provide a prosthesis which completely covers the defect, inconveniencing the patient no more than would an ordinary full upper dental plate.

After the anterior aspect of the tumor has been exposed by an adequate trans-maxillary approach, the growth must be further mobilized from its attachment at the base. If it were possible to dissect the mass from the underlying bone, periosteum, or fascia, it would then be feasible to enucleate the tumor in its entirety. Due to the irregular bony walls of the nasopharyngeal cavity, however, in addition to the generally broad dense attachment of nasopharyngeal fibroma, complete surgical removal of the neoplasm cannot be effected, unless it is limited to the basilar portion of the occipital bone where the surface is smooth. Residual tumor, therefore, will almost always be left behind, despite the good intentions of the surgeon. In many cases appreciable recurrences, frequently ulcerated and infected, will appear and have to be treated with interstitial radiation.

The indication for any given method of surgical removal must be decided by the clinical setting in each case. The selection of the proper time for operation may also be important. In general, we suggest deferment of any surgery in cases without marked deformity, hemorrhage, or sepsis. Such cases should be continued indefinitely under sex hormone and radiation therapy. Where hemorrhage is a serious complication or where surgical removal of the tumor is contemplated, a bilateral ligation of the external carotid arteries should first be made, hormones given, and radiation therapy instituted. By these less

aggressive measures, the hemorrhages may be arrested and the tumor reduced in size or at least brought under control. In any case, these preliminary precautions will markedly reduce the amount of bleeding at operation, if this finally becomes necessary.

TABLE I.—*Juvenile Nasopharyngeal Fibroma*

Tabulation of Methods of Treatment and Their Complications

Method of Treatment	Number of Cases	Case Number	Complications
A. X radiation	9	4	Dryness of nasopharynx
		8**	Atrophy of maxilae
		9**	None
		11	Dryness of nasopharynx
		12	None
		13**	None
		15**	Otitis media; coarse features
		19**	Otitis media
		*20**	Died of rheumatic fever
B. Interstitial radiation	5	3	Necrosis of palate
		5	Dryness of nasopharynx
		*7	Pansinusitis
		21	None
		29**	None
C. Interstitial and X radiation	2	2	Dryness of nasopharynx
		6	Dryness of nasopharynx
D. Surgical excision	4	*22	None
		*24	None
		*25**	None
		*26	None
E. Surgical excision combined with X radiation	1	*27	None
F. Surgical excision combined with interstitial radiation	2	1	Died of brain abscess
		*28	None
G. Surgical excision combined with interstitial and X radiation	4	*10	Died of brain abscess
		*16**	Necrosis of hard palate
		*17	Otitis media
		*18**	None
H. Hormones only	1	14	None
I. No treatment, observation only	1	23	None

* Ligation of external carotid arteries.
** Received hormone therapy.

METHODS OF TREATMENT SUMMARIZED AND COMPARED

Once the diagnosis of juvenile nasopharyngeal fibroma has been established, a therapeutic program should be adopted, subject to change as required by ensuing clinical developments. As already mentioned, there is no one type of treatment which is a panacea for any given case and in practically all instances a combination of methods must be employed. Obviously, aside from

periodic examination, an active therapeutic regimen is not indicated for the occasional case where the tumor is discovered during routine physical examination with no symptoms referable to it. In the absence of nasal obstruction, nose-bleeds, impairment of hearing, infection of the paranasal sinuses or middle ear, facial deformity, exophthalmos, headache or any discomfort about the head, or evidence of progressive enlargement or significantly increased vascularity of the neoplasm, a policy of watchful waiting is justified. This policy was followed in the case of one of our patients, age 15 on admission, and during a two-year period of observation the growth has remained stationary and asymptomatic.

Endocrine therapy was the only form of treatment which was prescribed in one case. In ten others it was combined with some form of radiation therapy, surgery, or both. With the combination of methods, sex hormone therapy was either utilized alone as the initial treatment and, following its failure to control the growth satisfactorily, radiation therapy was instituted, or both forms of treatment were begun simultaneously. While no definite regression of the tumor was noted in any case in which the patient received only male sex hormone, it was our clinical impression that hemorrhages usually diminished in frequency and severity following the use of hormones. The basis for this observation may be due to the considerations included under etiology.

In our series, some form of radiation therapy, either alone or combined with one or more surgical procedures, was resorted to in 23 cases (Table I). In nine of these, roentgen therapy was the only form of radiation employed and in this group late complications (dryness of nasopharynx, atrophy of maxillae, coarsening of features), attributable to the high dosages used to control the tumor, were frequent, indicating that no one method of treatment can be depended on without courting the hazards of over-aggressiveness. Because of its possible effect on the growth centers of the facial bones, roentgenradiation should preferably be reserved for patients 18 years of age or older.

Interstitial radiation therapy was employed alone in five cases and was followed by serious complications in three instances. In one, necrosis of the hard palate resulted, due to an unnecessarily large dose of gold seeds (total of 34 mcs. inserted at three sittings over a period of two years). In two, even more unfortunate developments followed massive doses of interstitial radiation (total dose of 32 mc. in one; 20 mc. in another after administration of 3000 r of 200 K.V. therapy through a single 6 cm. cheek portal). Both patients developed extensive osteonecrosis extending into the sphenoid bone and finally succumbed to brain abscess. At that time, about 20 years ago, we believed it essential to eradicate the growth completely in order to cure the patient and were not cognizant of the fact that simple control of tumor activity until sexual maturity had been attained was all that was necessary.

From an analysis of our data, it is noteworthy that as a rule smaller doses of interstitial and roentgenradiation will effectively control symptoms if the patient is receiving continued and adequate endocrine therapy, that bulky

tumors producing facial deformity or exophthalmos are followed by no complications when treated by primary surgical excision (four cases), and that residual tumors can be readily controlled with small doses of gold seeds inserted at frequent intervals.

During the period of radiation therapy, suppuration in the nasal accessory sinuses or middle ear occasionally supervenes and rarely mastoiditis may occur. Such sepsis, together with the untoward systemic effect of radiation therapy in general, makes hospitalization for the treatment of many of these patients mandatory. In this way local hygienic care, adequate nutritional therapy, blood transfusions, and chemotherapy are made available.

PROGNOSIS

As has been previously stated, if complications of hemorrhage, sepsis (pansinusitis, otitis media, etc.), and facial deformity by an expanding tumor can be prevented or even markedly reduced, little or no harm can come to the patient with nasopharyngeal fibroma and the growth will regress spontaneously in practically all instances. From a study of our cases it is plain that many of the most serious complications are brought on primarily by aggressive and ill-conceived therapeutic measures, such as poorly planned attempts at surgical removal without preliminary arterial ligation and inadequate exposure of the nasopharynx and secondarily by unduly vigorous endeavors to control further growth by radiation therapy. In such cases the patients would have been much better off without any treatment whatever.

Juvenile nasopharyngeal fibroma is not essentially a malignant tumor. It does not invade adjacent tissues and its destructive capacities are entirely due to pressure atrophy of contiguous structures by the enlarging mass. So far as we know, there is no well authenticated instance of malignant transformation of this tumor. In one case of the present series, the patient developed numerous recurrences, one of which showed on microscopic examination unusual cellularity and localized areas of malignant transformation. Seven subsequent recurrences were examined histologically, however, and none of these revealed any unsual changes. During the three years that have elapsed since the suspected malignant recurrence was noted, the tumor has not shown any evidence clinically of malignant propensities. Although Shaheen[26] stated that some of his cases underwent "carcinomatous and sarcomatous" changes, as mentioned earlier in this discussion, his series was not critically selected and in our opinion his conclusions are therefore unacceptable. We suspect that Shaheen's cases of "malignant transformation" were actually malignant nasopharyngeal cancers and not nasopharyngeal fibromas in the beginning. Two isolated case reports of malignant nasopharyngeal fibroma appeared in the literature in 1904 and 1912[14, 5] but here again the presented evidence is entirely unconvincing. Some of these errors undoubtedly arose from a mistaken morphologic interpretation of a highly vascular and highly cellular tumor as "angiosarcoma." Although the tumor in Wirth's patient[28] in the beginning simulated nasopharyngeal angiofibroma in some respects, the predominant

picture was that of chondrosarcoma, and it metastasized as such; since this case is complex and at complete variance with the accepted criteria necessary for a diagnosis of juvenile nasopharyngeal angiofibroma, it would hardly be reasonable to classify it as such.

In brief, the prognosis in a case of juvenile nasopharyngeal fibroma properly managed is excellent so far as the question of life is concerned. The greatest hazard both as regards life and permanent disability lies in injudicious treatment either by radiation therapy or surgery. As we have pointed out, the growth is practically incurable from the standpoint of its complete eradication and prevention of recurrences before sexual maturity is attained. After sexual maturity has been reached, however, the growth will disappear spontaneously in most instances.

END-RESULTS

Practically all patients with nasopharyngeal fibroma should recover and become symptom free when sexual maturity is reached. In our series there were two deaths due to brain abscess, resulting from over-dosage with interstitial radiation. A third patient succumbed to acute rheumatic fever just after the onset of treatment for nasopharyngeal fibroma.

SUMMARY

The thesis has been advanced that juvenile nasopharyngeal fibroma occurs only in pubescent males. For other reasons, namely its spontaneous or readily induced regression with the appearance of full sexual development, a sex-endocrine relationship for this tumor probably exists. Although its histogenesis has not been definitely established, a vascular origin is proposed in this report. This proposal is based on specific involutional changes which can be induced with male sex hormone therapy and irradiation. The induction of vascular changes by hormone administration has considerable experimental support.

Even though nasopharyngeal fibroma is essentially benign, it frequently produces serious and disabling symptoms because of its progressive growth and tendency to profuse hemorrhage. The indications, methods, and hazards of both radiation and surgical treatment in combating these symptoms are discussed. The trend on the Head and Neck Service at Memorial Hospital has been to employ one or more of these methods of treatment in an effort to control the growth since, in most cases, spontaneous regression can be expected after sexual maturity. Aggressive measures either by irradiation or surgery will never completely and permanently eradicate the neoplasm and will involve considerable risk of ultimate disability or even death.

REFERENCES

[1] Allen, Edgar: Sex and Internal Secretions. A Survey of Recent Research, ed. 2, Baltimore, Williams & Wilkins, 1939.

[2] Bensch, H.: Beitrage zur Beurtheilung der Chirurgischen Behandlung der Nasenrachenopolypen, Breslau, E. Morgenstern, 1878.

[3] Chaveau, C.: Histoire des Maladies du Pharynx, Paris. J. B. Bailliere et Fils, vol. 5, 1906.

[4] Chelius, J. M.: System of Surgery, vol. 2 (translated by John F. South). London, Henry Renshaw, 1847.

[5] Dabney, V.: Fibroma of the Nose and Nasopharynx with Sudden Malignant Degeneration. Laryngoscope, **22**: 960–964, 1912.

[6] Delevan, D. B.: The Effect of Radioactivity upon Nasopharyngeal Fibromas. Med. Rec., 1915.

[7] Edwards, E. A., J. B. Hamilton, S. Q. Duntley, and G. Hubert: Cutaneous Vascular and Pigmentary Changes in Castrate and Eunnuchoid Men. Endocr., **28**: 119–128, 1941.

[8] Ewing, J.: Neoplastic Diseases, ed. 4, Philadelphia, W. B. Saunders Co., 1941.

[9] Figi, F.: Fibromas of Nasopharynx. J. A. M. A., **115**: 665–671, 1940.

[10] Goldsmith, P. G.: Nasopharyngeal Fibroma; a Clinical and Pathological Study. J. Laryngol. & Otol., **38**: 565–574, 1923.

[11] Gosselin, Paul: Clinique Chirurgicale de l'hôpital de la Charité, ed. 2, Paris, 1876.

[12] Hamilton, J. B.: Changes in Nasal Mucosa of Monkeys (Macaca rhesi) and Humans by Male Hormone Substances. Proc. Soc. Exper. Biol. & Med., **37**: 366–369, 1937.

[13] Hippocrates: Oeuvres Completes, Traduction Nouvelle par E. Littre, Paris, J. B. Bailliere, 1839–1861, 10v.

[14] Jackson, C. D.: Fibroma of the Nasopharynx, Tracheotomy, External Carotid Ligation, Extirpation, Laryngoscope, **14**: 267–270, 1904.

[15] Jackson, C., and G. M. Coates: The Nose and Throat and Ear and Their Diseases. Philadelphia, W. B. Saunders Co., 1930.

[16] Langenbeck, C. J. M.: Nosologie und Therapie der Chirurgischen Krankheiten, Göttingen, vol. 5, 1834.

[17] Legouest, C.: Proceedings of Surgical Society of Paris, 1865.

[18] McGrath, E. J., and L. G. Herrmann: Influence of Estrogens on the Peripheral Vasomotor Mechanism. Ann. Surg., **120**: 607–615, 1944.

[19] Mortimer, H., R. P. Wright and J. B. Collip: Effect of Administration of Oestrogenic Hormones on Nasal Mucosa of Monkey (Macaca mulatta). Canad. M. A. J., **35**: 503–513, 1936.

[20] *Idem:* Effect of Oestrogenic Hormones on Nasal Mucosa; Their Role in Naso-Sexual Relationship; and Their Significance in Clinical Rhinology. Canad. M. A. J., **335**: 615–621, 1936.

[21] New, G. B., and F. A. Figi: Treatment of Fibromas of the Nasopharynx: Report of 32 Cases. Am. J. Roentgenol., **122**: 340–343, 1924.

[22] Ollier Quoted by Nicholas Senn: The Pathology and Surgical Treatment of Tumours, ed. 2, Philadelphia, W. B. Saunders & Company, 1900.

[23] Reynolds, S. R. M., and F. I. Foster: Specific Differences in Cholinergic Action of Estrogen. Am. J. Physiol., **131**: 200–202, 1940.

[24] Reynolds, S. R. M., J. B. Hamilton, J. R. DiPalma, and G. Hubert: Dermovascular Actions of Certain Steroid Hormones in Castrate, Eunnuchoid and Normal Men. J. Clin. Endocr., **2**: 228–236, 1942.

[25] Rouge: Quoted by Nicholas Senn: The Pathology and Surgical Treatment of Tumours, ed. 2, Philadelphia, W. B. Saunders & Company, 1900.

[26] Shaheen, H. B.: Nasopharyngeal Fibroma. J. Laryng. and Otol., **45**: 259–264, 1930.

[27] Soskin, S., and L. B. Bernheimer: Mechanism of the Estrogen Effect on the Nasal Mucosa in Atrophic Rhinitis. Proc. Soc. Exper. Biol. & Med., **42**: 223–224, 1939.

[28] Wirth, J. E., and M. B. Shimkin: Chondrosarcoma of the Nasopharynx Simulating Juvenile Nasopharyngeal Fibroma. Arch. Path., **36**: 85–88, 1943.

[29] Verneuil: Quoted by H. Bensch: Beitrage zur Beurtheilung der Chirurgischen Behandlung der Nasenrachenpolypen. Breslau, E. Morgenstern, 1878.

TRANSPERITONEAL APPROACH TO THE INTERVERTEBRAL DISC IN THE LUMBAR AREA

JOHN D. LANE, JR., M.D., F.A.C.S., SENIOR SURGEON
AND
EMORY S. MOORE, JR., M.D., SENIOR ASSISTANT SURGEON

DEPARTMENT OF SURGERY, U. S. MARINE HOSPITAL, BALTIMORE, MARYLAND
HOSPITAL DIVISION, UNITED STATES PUBLIC HEALTH SERVICE

THE SYNDROME OF LOW BACK PAIN, with various sciatic nerve symptoms, is now a well recognized entity and constitutes one of the major groups of chronic disabling conditions occurring in individuals during the most productive years of life. One of the etiologic conditions has now been scientifically established; that it is no longer a hypothesis has been demonstrated by sound research work in the past 10 years. Statistics indicate that alterations in the structure and contour of the intervertebral disc at the 4th and 5th lumbar spaces are responsible for a large percentage of disabling conditions in the low back. Although the etiology and pathology are well recognized, the treatment remains a very controversial subject. The basis of this is the lack of uniformly good results.

The purpose of this publication is to present a technic fashioned to improve upon some of the previously known principles thought to be prime factors in obtaining good results in the treatment of degenerative changes in intervertebral discs. It is not intended in this presentation to offer any contributions or criticisms of present methods of diagnosis or other clinical relations, except in so far as they may be connected with the present surgical technic described.

Poppen[1] in a review of 400 postoperative disc cases noted perfect results in only 38 per cent, the remainder having varying degrees of disability classed as undesirable. Kirstein[2] reports 72 per cent of his operated cases as free from "sciatica," 48 per cent completely free from local back symptoms, and only 24 per cent as symptom-free. DeBakey reports on 2,450 cases of ruptured intervertebral discs in the U. S. Army to the date of August, 1944. Analysis of these cases reveals that 75 per cent of the group were treated without surgery; 78 per cent of this conservatively treated group were discharged because of this condition. Of the group that received surgery, 31 per cent were discharged during the first year after surgery. In a previous article published by one of us,[3] a series of 136 operated cases and 193 cases conservatively treated showed complete relief was obtained in only 56 per cent of the entire group.

It is now evident that the symptoms resulting from degeneration and injury to the intervertebral disc are rather easily diagnosed, but the present form of treatment leaves much to be desired as demonstrated by the above statistics. Too many cases are not sufficiently improved or have a return of symptoms at a later date. To improve these too frequent failures in treatment,

a surgical procedure dealing with the diseased disc has been devised, using the transperitoneal approach with ox bone implantation. The objective, anatomic and physiologic principles, operative treatment, and follow-up statistics are presented.

The object of this procedure is:

1. To completely remove the entire diseased disc with the cartilaginous end plates of the adjacent vertebrae.

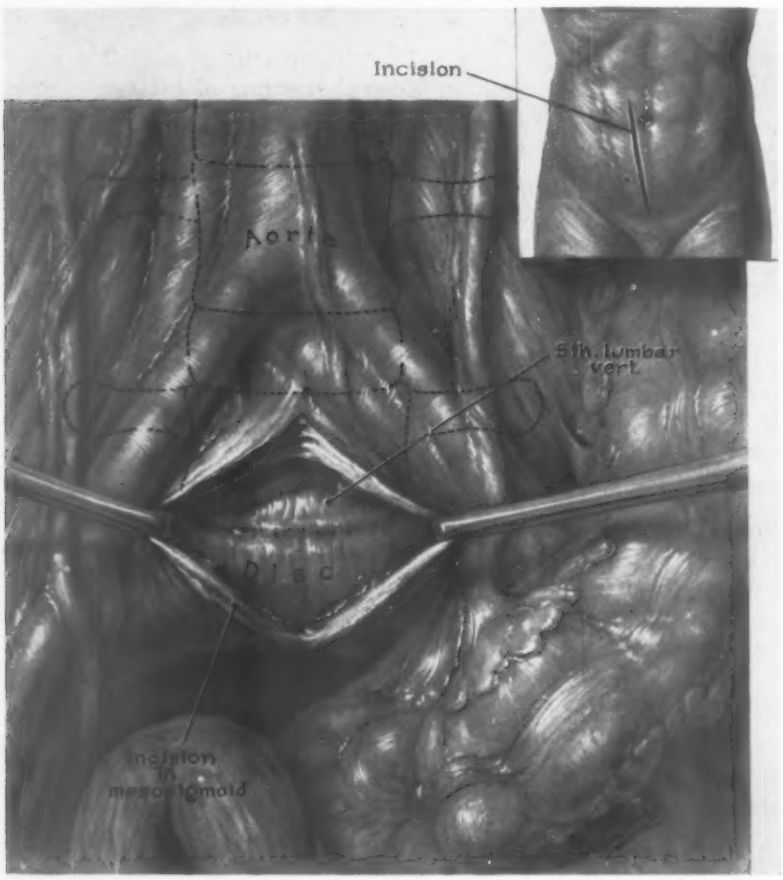


FIG. 1.—Demonstrates abdominal incision. Important pelvic structures, and location of 5th lumbar disc.

2. To wedge the disc space open with an ox bone implantation in order to maintain normal space between the vertebrae until firm bony fusion is obtained.

There are several undesirable features in the posterior approach by partial laminectomy, which have contributed in some degree to the large number of unsatisfactory results. The usual approach by hemilaminectomy fre-

quently gives an inadequate exposure, so that only a small area of the disc space is visualized on the posterolateral side on which the laminectomy is performed. If the herniation of the diseased disc is in the intervertebral canal more anteriorly, it may not be visualized. In exposing the disc, retraction of the nerve roots may result in permanent damage or irritation to these structures. Hemorrhage from the anterior longitudinal veins often prevents

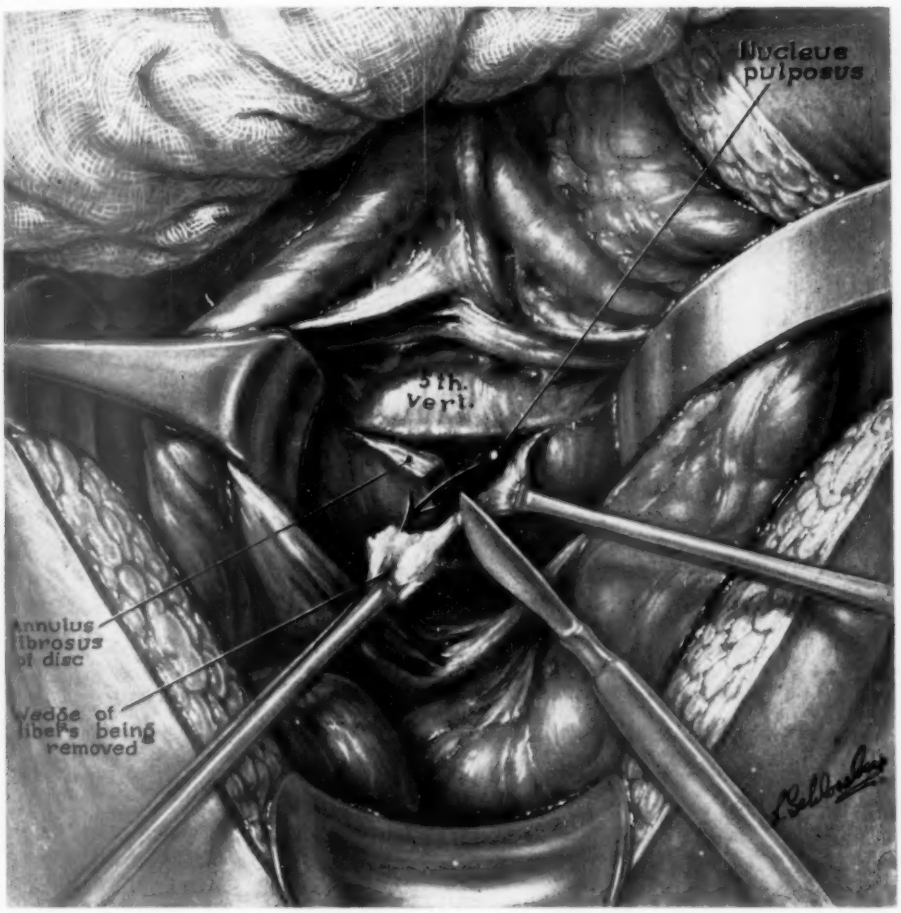


FIG. 2.—Method of opening the anterior longitudinal ligament and adequately exposing the disc space and nucleus pulposus.

adequate vision in demonstrating the lesion and has been found to be a factor in causing postoperative sequelae by hematoma formation followed by fibrosis and nerve root irritation with dural adhesions. Only a small portion of the disc and cartilaginous end plates can be removed by the posterior approach. Since the annulus fibrosus has been damaged, more trauma to the disc can

produce a further herniation of the remaining nuclear material, with a subsequent return of symptoms. Due to removal of the nuclear material, there is a tendency, first, toward subsequent narrowing of the disc space with resulting imbalance of weight bearing between the vertebral bodies and the corresponding facets and, second, joint instability. We have not obtained the desirable features of bone graft fusion and normal disc space by the posterior approach.

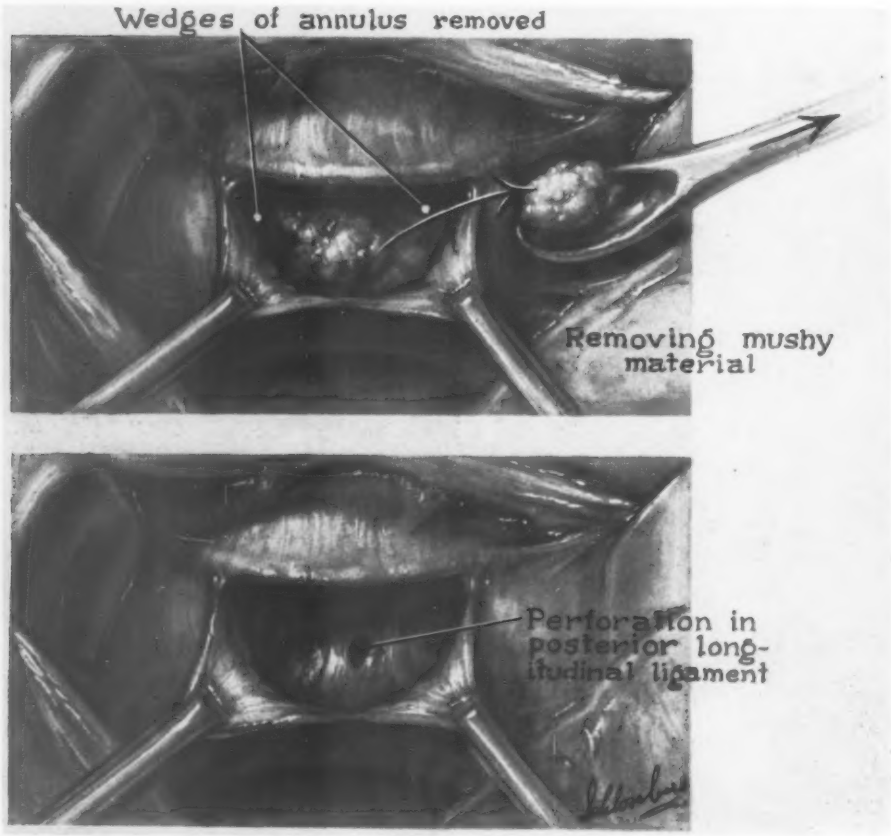


FIG. 3.—(A) Removal of all material within the disc cavity.

(B) Shows exposure of posterior longitudinal ligament with defect after complete removal of disc contents.

In removing diseased discs, by the anterior transperitoneal route, the following results, which we believe beneficial, have been obtained. (1) Good exposure to the entire disc space and cartilaginous end plates. (2) The 3rd, 4th, and 5th discs can be examined and treated through the same abdominal incision. (3) Removal of the entire disc and all cartilaginous end plates, suffi-

cient to obtain good bony union, can be accomplished. (4) Hemorrhage is easily controlled and does not occur into the spinal canal, and no trauma or retraction of the cord or nerve roots are necessary. (5) A large bone implantation can be wedged into the disc space to prevent narrowing until solid bony fusion between the adjacent vertebral bodies has taken place. These are major factors in determining a recovery from symptoms of degenerated disc.

Some knowledge of anatomy of the abdominal cavity is necessary in carrying out the technic of the transperitoneal approach. The blood supply and mesenteric attachments of the lower ileum and colon, the relationship of the pelvic portions of the lower ureter to the spine and great vessels, and the relationship of the lower vena cava and common iliac vessels to the vertebral bodies should be thoroughly understood (Fig. 1).

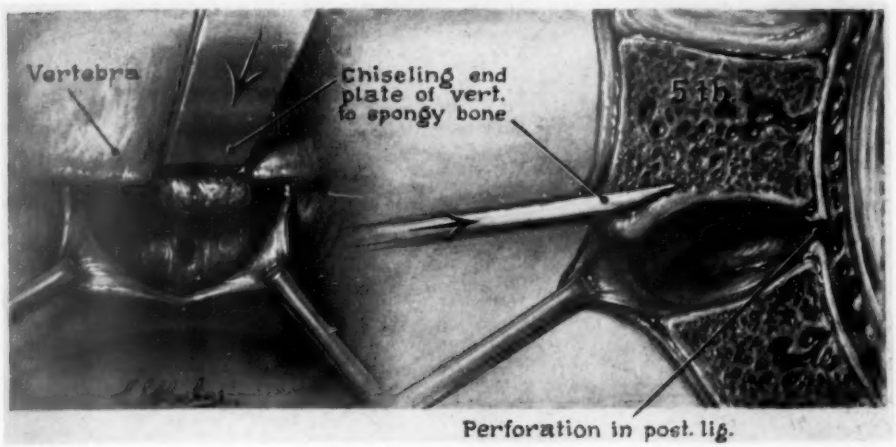


FIG. 4.—(A) Anterior view demonstrating removal of cartilaginous end plates exposing cancelous bone to promote fusion.

(B) Same procedure in cross section.

SURGICAL PROCEDURE

In carrying out this procedure, the incision is paramedian from the symphysis to 3 inches above the umbilicus (Fig. 1). The rectus sheath is incised 1 inch lateral to the midline, rectus muscle retracted laterally, peritoneum incised, and the abdomen explored for any pathologic condition. Slight Trendelenburg position is obtained to keep the intestines in the upper abdomen. The redundant part of the sigmoid, cecum, and small gut is then displaced in the upper portion of the abdomen and maintained with moist laparotomy sponges to give good exposure to the lower lumbar and pelvic portion of the posterior peritoneum. The pelvic portion of the colon is then retracted to the left, after identifying the ureters. An incision is made in the posterior pelvic peritoneum in the midline, beginning over the sacrum and

extending to the bifurcation of the aorta (Fig. 1). The kidney bar on the operating table, previously placed beneath the 4th lumbar vertebra is now elevated sufficiently to push the lumbar spine into hyperextension and forward into the abdominal cavity. This gives better exposure and makes as wide as possible the intervertebral disc space in the lower lumbar spine, considerably increasing the access to the space to be opened. From 1–2 inches below the

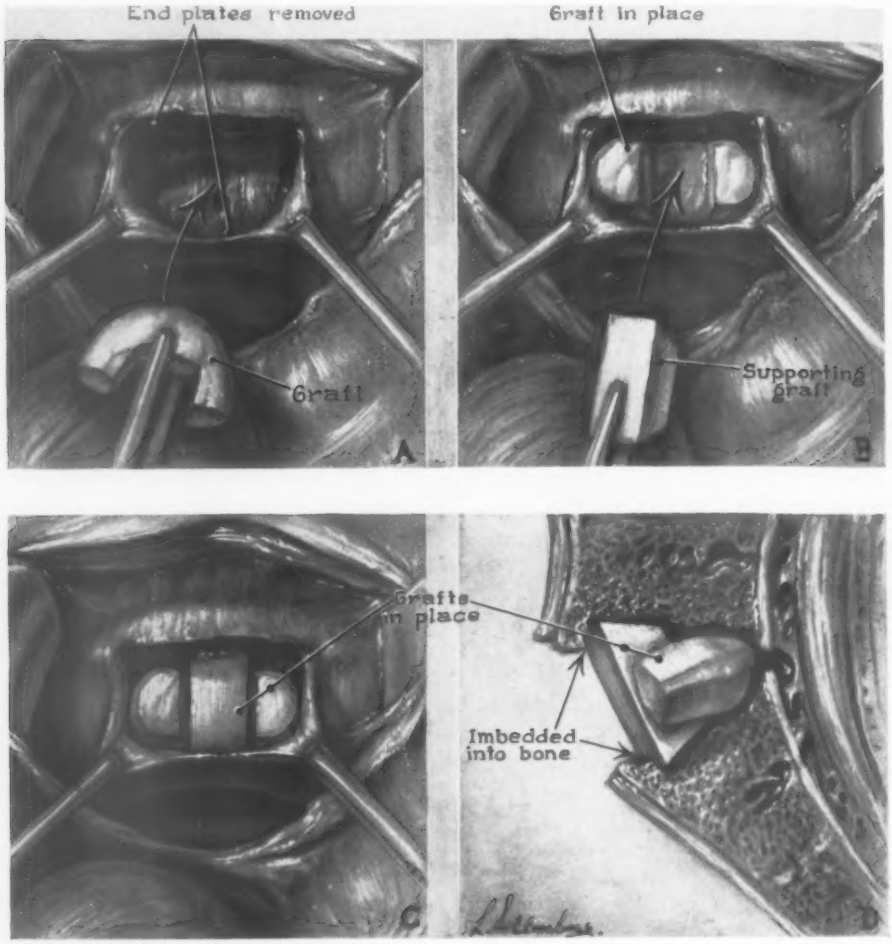


FIG. 5.—Method of inserting ox bone crescent and peg in disc space.

bifurcation of the aorta, the 5th disc is located by palpation between the common iliac vessels and is distinguished by the palpating finger by a distinct elevation with a rubbery consistency as contrasted to the hard vertebral bodies. The presacral sympathetic nerve plexus and veins are freed by blunt dissection and retracted to one side, thus completely visualizing the anterior longitudinal ligament over the prominence of the 5th lumbar disc. The disc can then be

examined for consistency. By lowering the kidney bar to reduce the lordosis, an estimate of disc narrowing is obtained.

To enter the disc space the kidney bar is re-elevated and a transverse incision made across the anterior longitudinal ligament at the lower margin of the 5th vertebra sufficient to give access to the entire anterior disc space. A vertical incision is made from the mid portion of the transverse incision to the upper margin of the sacrum. This allows for the turning back of a flap of the anterior ligament to give access to the entire nuclear material (Fig. 2). The state of the nuclear material can then be easily ascertained. The contents of the disc space are easily removed by a curette, with a deep cup, and a sharp cutting edge, which facilitates the cutting away of the cartilaginous end

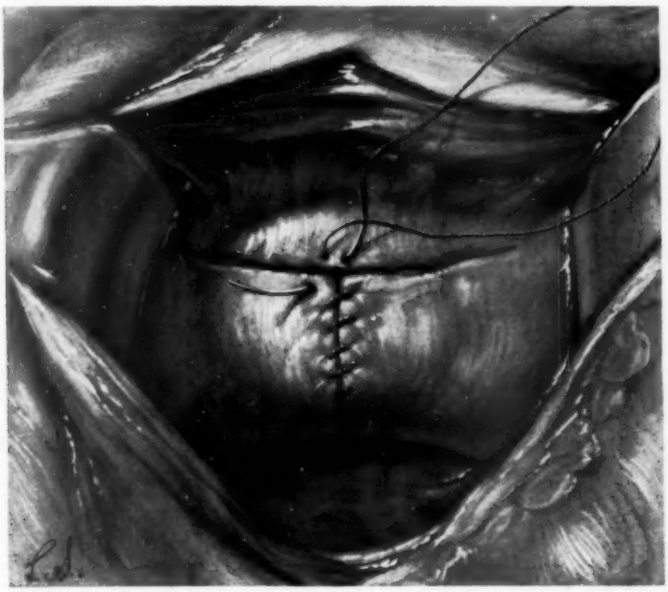


FIG. 6.—Closure of disc space by suture of anterior longitudinal ligament

plates, as well as the nuclear material. It is necessary for the handles of the curette to be about 8 inches in length, with a cross bar at the proximal end to get sufficient leverage and motion to cut away the bands of the annulus fibrosus and cartilage. The disc contents are removed until the ligaments retaining the disc are visible around the entire disc space (Fig. 3, A & B). Often there is relaxation and bulging of the ligament outward, due to previous degeneration of the disc material, allowing the disc space to narrow and bulge. This outward bulging may be most prominent on either side or posteriorly, and it is always more prominent in these locations than anteriorly since the annuli fibrosi are stronger and several times thicker anteriorly than on the sides or posteriorly. The ligaments at the posterior and lateral sides are then

thoroughly explored with a small blunt instrument to indicate any weakness or defective space at the site where preoperative clinical findings have suggested nerve root pressure. If defects or openings are found, they are further spread open to ascertain if a portion of the nuclear material has been extruded through the defect (Fig. 3B). Such material, if found, is removed. After sufficient search has been made to determine that all nuclear material has been excised, the cartilaginous end plates are completely removed from the surface of the vertebrae by a sharp curette or chisel (Fig. 4, A & B). This procedure is done last, as it may cause considerable oozing of blood. If done before a thorough search of the disc has been made, it may obscure a defect in the ligament or a portion of nuclear material in the lateral gutter. If bleeding is too free following removal of the end plates, a tight packing with a gauze sponge for several minutes will usually suffice to control it.

To maintain the disc space in its normal width while fusion is progressing, a specially prepared ox bone wedge is used. This was selected on the basis of surgical work previously reported by Orell[5] working on fractures, in which he demonstrated that ox bone heterogenous implantations made an excellent bridge and were slowly absorbed over a period of from 12–20 months, gradually becoming a spongy mass through which new capillaries and osteoblasts could permeate and form new bone. This course of events permits gradual replacement of the ox bone wedge by homologous bone from the adjacent vertebral bodies and leaves less chance for narrowing during the fusion process. The ox bone wedge now used consists of a crescent shaped piece with beveled edges, which is driven into the posterior portion of the disc space with the spine hyperextended (Fig. 5B). Between the wings of the crescent wedge, a large square bone peg of the same material is driven, being previously measured to fit just inside the wings of the crescent (Fig. 5C). This almost completely fills the disc space. Following wedging of the disc space, the flaps of the anterior ligament are closed and sutured over the disc space in their normal position, thus completely encasing the ox bone within the disc space (Fig. 6). The kidney rest is lowered, reducing the hyperextension of the spine and spreading the disc space open wider by impinging on the ox bone wedge.

Exposure of the 3rd and 4th disc spaces is slightly more technical than that of the 5th. However, complete visualization can be accomplished. This is due to the fact that the 5th disc lies below the bifurcation of the great vessels, whereas, the 3rd and 4th are beneath the aorta and vena cava. Exposure of the 3rd and 4th is readily achieved for examination, and it is always done if there is any clinical evidence of nerve root irritation or joint instability on examination previous to surgery. This is done by extension upward on the posterior peritoneal incision over the iliac vessels either to the right or left of the midline and along the lateral border of the spine, sufficiently to expose the 3rd and 4th discs. If the incision is on the right side, the ureter is retracted outward and the loose areolar tissue gently dissected through until the lateral border of the vena cava is visualized. A short bladed right

angle blunt retractor is placed beneath the lateral border of the vena cava, and with gentle retraction it is displaced to the left along with the aorta, until the disc space is adequately exposed. If exposure is done from a left sided approach instead of the right, the incision is the same as that of the right, except that the lateral border of the aorta is encountered first instead of the vena cava. In a like manner the aorta is freed at its lateral border and is retracted toward the right side, with the vena cava, until adequate exposure

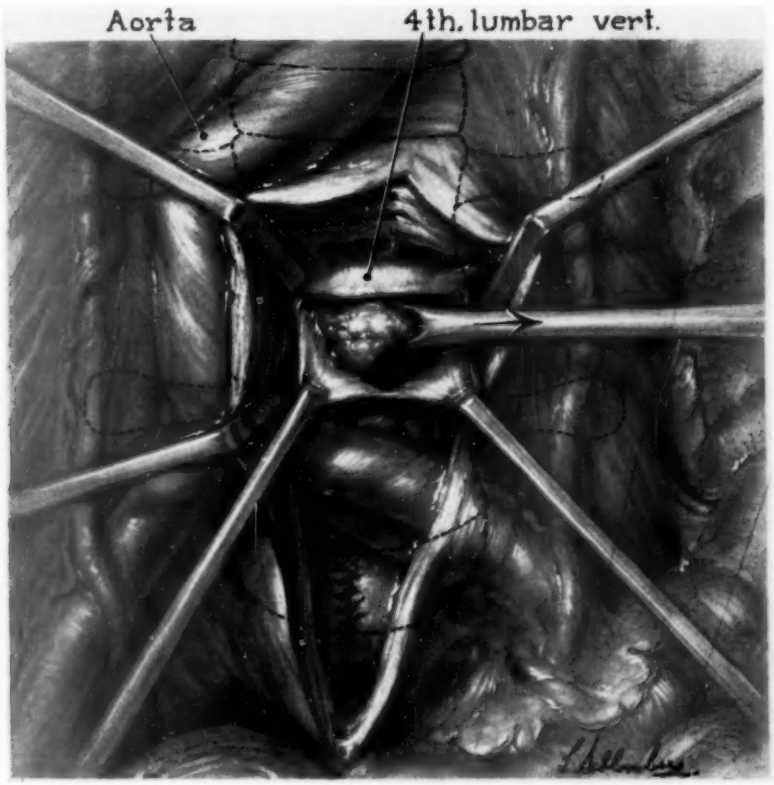


FIG. 7.—Method used in exposing the 3rd or 4th disc spaces.

of the disc space is obtained from this side (Fig. 7). If deemed necessary, this disc space may be treated in a similar manner as already described for the 5th. Closure, following the completion of this procedure, consists of suture of the anterior longitudinal ligament to its adjacent vertebra, completely closing the intervertebral space. This is accomplished with sutures of cotton No. 30. Closure of the posterior and anterior peritoneal opening is obtained with sutures of interrupted cotton No. 70. Fascial layers of the anterior and posterior rectus sheaths are approximated with interrupted sutures of cotton No. 30.

POSTOPERATIVE CARE

Postoperative care has been most conservative, in order to arrive at a definite conclusion as to the ideal convalescence which will not interfere with fusion. Measures to combat gaseous distention and phlebitis are followed. The patients are placed in bed with firm rigid support beneath the mattress. No braces or casts are applied at this time. The patients are kept in a supine position on a hard surface for 30 days. X-rays of the lumbar spine are then taken and a body cast is applied to cover the entire lumbar spine and sacrum.

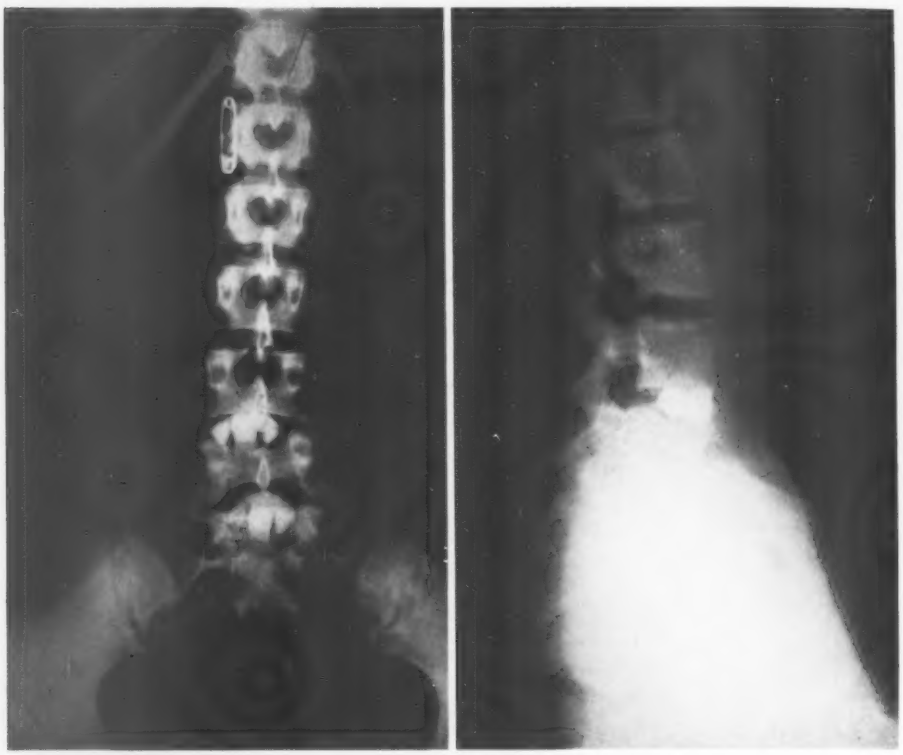


FIG. 8.—(a) (Left) AP X-ray for position of bone implantations 30 days postoperative. (a) (Right) Lat X-ray for position of bone implantation 30 days postoperative.

The lumbar spine is held in slight hyperextension. Some patients have insisted on being out of bed on the 15th postoperative day because they have felt normal. Against our advice, this was done by three patients without support, and they developed no back complaints. The patients are allowed to be ambulant and to return to their homes after the application of the cast and examination, including roentgenograms, for evaluation of progress. If convalescence is satisfactory, they are then supplied with a lumbosacral belt and instructed to refrain from strenuous use of the back; they return at monthly intervals for further check-up examination, including physical examination, check on symptoms, and roentgenograms, to determine the progress of fusion.

This procedure was first started at the Marine Hospital on February 27, 1946, and to the present date all discs requiring surgery have been treated in this manner.

CLINICAL MATERIAL

A diagnosis of herniated nucleus pulposus was made in 97 cases admitted to this hospital between February 1, 1946, and February 1, 1947. All patients were treated conservatively by peridural block, traction, or plaster body cast, or a combination of these until such treatment was found ineffectual in the individual case. Thirty-six cases either failed to respond to such treatment or their improvement was insufficient to allow return to a gainful occupation. These 36 cases were subjected to surgery and form the basis of this report.

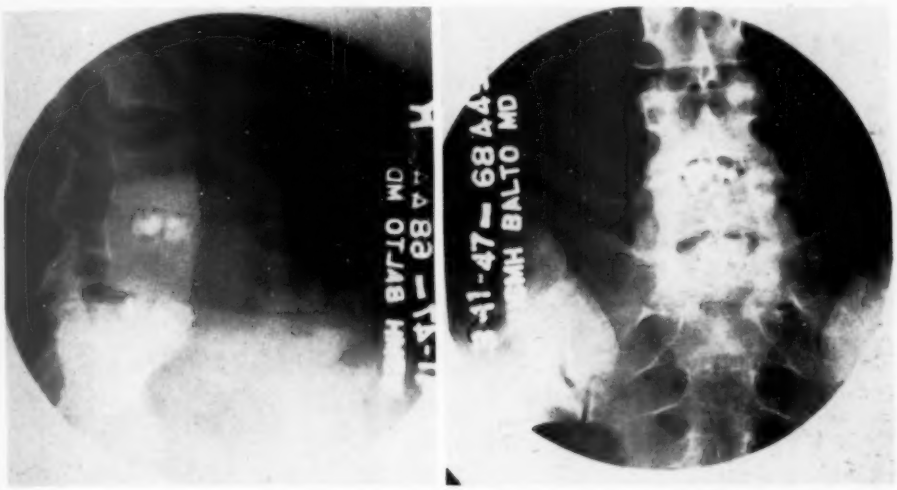


FIG. 8.—(b) (Left) AP X-ray for progress 5 mos. postoperative. Fusion present. (b) (Right) Lat X-ray for progress 5 mos. postoperative. Fusion present.

Of the patients undergoing surgery, 29 were males and 7 females. The average age was 31.9 years—the youngest 18 years and the oldest 52. Twenty-nine of the patients followed manual occupations; 7 held sedentary positions.

The average duration of symptoms at the time of surgery was 19.7 months. Of the 36 cases undergoing surgery; 1 case had sciatic neuritis alone, 3 cases had back complaints and findings without neurological changes in the lower extremities, and the remaining 32 cases had back and lower extremity symptoms and neurological findings.

Roentgen examination of the lumbar spine (A-P and lateral) revealed: Narrowing of the suspected space in 8 cases; evidence of previous partial hemilaminectomy in 4 cases; spondylolysis in 3 cases; spondylolisthesis in 1 case; congenital defect of the lamina in 1 case; and lumbarization of the 1st sacral vertebra in 1 case. Pantopaque myelography appeared indicated in 7 patients to substantiate further the diagnosis of herniated nucleus pulposus

or to rule out intravertebral canal pathology. The test was suggestive of the former in 3 patients and negative in 4.

FINDINGS AT SURGERY

At the time of operation, 25 of the 36 cases (69.4 per cent) had single disc lesions as follows: Lumbar IV—3 patients; Lumbar V—22 patients. Eleven of the 36 cases (30.6 per cent) had multiple disc lesions as follows: Lumbar III and V—1 patient, Lumbar IV and V—9 cases; Lumbar V and VI—1 case.

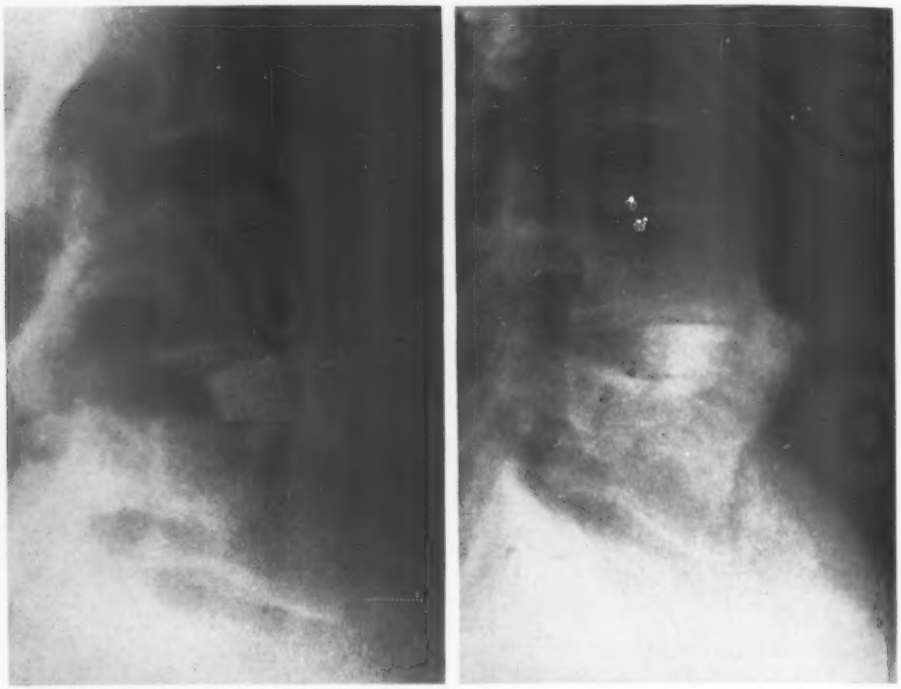


FIG. 9.—(a) Lat X-ray for position of bone implantation 30 days postoperative. (b) Lat X-ray for progress 11 mos. postoperative. Fusion present.

POSTOPERATIVE COMPLICATIONS

Postoperative complications occurred in 5 patients, or 14 per cent, as follows: Phlebothrombosis (femoral vein) 2 cases (5.6 per cent); wound separation 1 case (2.8 per cent); hypostatic pneumonia 1 case (2.8 per cent); cystitis (acute) 1 case (2.8 per cent). Of the 2 cases of phlebothrombosis, 1 was of minor intensity and had no sequelae. The other patient continues to have minor residual swelling of the involved extremity. The postoperative wound separation occurred on the 11th postoperative day in a very obese and uncooperative patient. Evisceration did not occur. However, he developed a ventral hernia which has subsequently been repaired. The hypostatic pneumonia occurred on the 5th postoperative day and was controlled within 48

hours by chemotherapy. The patient had no sequelae. The complication of postoperative cystitis appeared on the 5th day of convalescence and was secondary to catheterization. Forced fluids and sulfadiazine controlled this complication within a period of three days.

RESULTS

Since the time of surgery, the patients have been followed as outlined previously. All cases, with one exception, have returned for monthly check-ups. This patient was last seen after his 4th postoperative month, at

TABLE I.

		Back Symptoms	Leg Symptoms	Neurologic Findings	General Status	
					Cases	Percent Total
Less than 4 months (10 cases)	Unchanged	2	1	1	1	2.9 per cent
	Improved	8	1	1	9	25.7 per cent
	Asymptomatic	0	8	8	0	
4–8 months (14 cases)	Unchanged	0	0	0	0	
	Improved	13	8	4	13	37.0 per cent
	Asymptomatic	1	7	10	1	2.9 per cent
8–12 months (11 cases)	Unchanged	1	1	0	1	2.9 per cent
	Improved	4	2	4	5	14.3 per cent
	Asymptomatic	6	8	7	5	14.3 per cent
	Total unchanged		5.8 per cent			
	Total improved		77.0 per cent			
	Total asymptomatic		17.2 per cent			

TABLE I.—Tabulation of symptoms and general status of cases in postoperative periods from 1 to 4 mos.—4 to 8 mos.—and 8 to 12 mos.

which time he was asymptomatic. Since he has subsequently been lost for follow-up examination, his case has been excluded from the result series.

Final results at the time of this writing are, of course, impossible to evaluate, since inadequate time has elapsed. Furthermore, the series is too small for statistical value. However, definite trends toward an ultimate result can be seen. For convenience of analysis the postoperative status of these cases has been divided into 4-month periods as follows:

Less than 4 months — 10 patients
4 to 8 months — 14 patients
8 to 12 months — 11 patients

Table I utilizes the above grouping and tabulates the status of the patients at the time of this writing. In brief, 2 or 5.8 per cent, of the 35 cases are unchanged, and 6, or 17.2 per cent, are asymptomatic. The remaining 27 cases, or 77 per cent, are improved. However, without exception in this improved group, residual symptoms and findings have decreased in intensity

and frequency in direct relationship to the length of time from the date of surgery. This observation tends to indicate that eventually these patients will be classified as symptom-free. Generally the chart demonstrates that this complete recovery is slow. This is to be expected, since advaiced bony fusion of the involved intervertebral joint is necessary before the patient becomes completely asymptomatic. This principle is substantiated in the roentgen-ray findings which are tabulated in Table 2. Those patients in whom there is demonstrated bony fusion by roentgen-ray are asymptomatic.

It has been found that bony callus appears in 3–4 months in some cases. Fusion is usually not seen by roentgenograms until the 8–12 month period.

TABLE II.—*X-ray Findings.*

Time Intervals	Callus Absent	Callus Present	Fusion Present
Less than 4 months (10 cases).....	5	5	0
4–8 months (14 cases).....	0	13	1
8–12 months.....	0	5	6
	5	23	7

TABLE II.—X-ray findings in postoperative cases indicating progress of fusion in the 4 mos. period of the 1st year.

At the present time one case demonstrates early fusion in the 4–8 month group (Fig. 8). One of the cases in the 8–12 month group demonstrates fusion and is illustrated in Fig. 9.

All patients have been advised most conservatively in regard to returning to duty or work. It has been our feeling that definite harm to the softened bone graft and early callus might be produced by too strenuous activity. Though many patients have felt capable of performing duty after three months of convalescence, we have not recommended their return to work until there is adequate evidence of early fusion or advanced callus formation. Table 3 tabulates the work status of the 35 cases. As may be seen in the table, these patients do not return to duty until after the 4th month of convalescence. Fourteen patients or 40 per cent have returned to duty.

At this time, one of the 8–12 month group is considered as unsatisfactory. The patient's symptoms of back pain have not shown enough improvement for any rehabilitation, the sciatic symptoms are improved, and callus formation is present on roentgen-ray examination.

This procedure has been devised as an attempt to improve on some of the undesirable features of the posterior approach and the surgical treatment of symptoms caused by degeneration of the intervertebral disc. It is realized that meticulous care must be used to exclude other diseases and intra-spinal lesions before carrying out this procedure; although it gives excellent exposure to the disc, it does not allow for complete exploration of the spinal canal and cord. Further, it is our opinion that all clinical symptoms and physical findings should be carefully analyzed to localize the nerve roots and

disc spaces before surgery, thus effecting a better evaluation of the findings at operation with the preoperative symptoms. It is realized that as small a series of cases as is now presented will not be sufficient to make a definite evaluation as to its efficiency in treating degenerated discs. Neither is the duration of time that has elapsed since surgery sufficient to allow a final conclusion on this small series. However, we believe there are certain aspects in the principles applied and trends in the beneficial results so far obtained which make this publication worth while.

TABLE III.—*Duty Status.*

Time Interval	Off Duty	Light Duty	Full Duty
Less than 4 months (10 cases).....	10	0	0
4–8 months (14 cases).....	9	3	2
8–12 months (11 cases).....	2	3	6
	—	–	–
	21	6	8
	(60 per cent)	(17.1 per cent)	(22.9 pe. cent)
	—	–	–
	21	6	8
	(60 per cent)	(17.1 per cent)	(22.9 per cent)

TABLE III.—Duty status of postoperative cases, arranged in 4 mos. periods during 1st year.

CONCLUSIONS

1. A method of complete removal of either the 3rd, 4th, or 5th lumbar intervertebral disc, with technic to fuse the joint, is presented.

2. The principles and technic are outlined.

3. This procedure has been used since February 27, 1946, in 36 cases of herniated intervertebral discs. Postoperative complications have occurred in five patients. There have been no deaths.

4. A preliminary survey of monthly follow-up studies on 35 of the 36 postoperative cases reveals six cases asymptomatic, 27 improved, and two unchanged. The course of improved cases is toward complete recovery from symptoms existing prior to surgery. Fourteen patients have resumed either light or regular duty.

5. The obliteration of the involved disc space by bony fusion is necessary for complete amelioration of symptoms.

BIBLIOGRAPHY

[1] Poppen, James L.: Herniated Intervertebral Disc: An Analysis of 400 verified cases. New England J. Med., **232**: 211,

[2] Kirstein, Lemart: Follow-up Examination of Operated and Non-operated Cases, with Clinical Symptoms of Herniated Disc Acta Med. Scandinav., **CXX**: 93, 1945.

[3] Lane, J. D.: Surgical Treatment of Intervertebral Discs. New Orleans M. & S. J., **97**: 270–275, 1944.

[4] Orell, Svante: Studies of Bone Implantation, New Bone Formation Implantation of Os Purum and Transplantation of Os Novum. Acta Chir. Scandinav., (Supp. 31) **74**: 1, 1934.

[5] Orell, Svante: Surgical Bone Grafting with Os Purum, Os Norum and Boiled Bone. J. Bone and Joint Surg., **19**: 873, 1937.

A SIMULTANEOUS ABDOMINAL AND PERINEAL APPROACH IN OPERATIONS FOR IMPERFORATE ANUS WITH ATRESIA OF THE RECTUM AND RECTOSIGMOID

J. E. Rhoads, M.D., R. L. Pipes, M.D., and J. Perlingieró Randall, M.D.

FROM THE DEPARTMENTS OF SURGERY AND PEDIATRICS, HOSPITAI OF THE UNIVERSITY OF PENNSYLVANIA AND THE DEPARTMENTS OF PEDIATRICS AND PATHOLOGY, CHILDREN'S HOSPITAL OF PHILADELPHA

There is a small group of patients with imperforate anus which presents difficult surgical problems because the large bowel is absent or has no lumen from a point within the peritoneal cavity to the anal dimple so that it cannot be reached satisfactorily from the perineum. These cases correspond to Type III of the Ladd and Gross[1] classification.

In two such patients in whom the sigmoid, as outlined by roentgen ray, stopped at a point about half-way between the anal dimple and the umbilicus, we have carried out a one-stage abdomino-perineal procedure working through both perineal and abdominal incisions at the same time. We believe the simultaneous approach has certain advantages which may help to make the one-stage operation feasible.

The mortality in the two-stage procedure, as reported by five authors,[1, 2, 3, 4, 5] has been discouragingly high, and in the single case in which a one-stage abdomino-perineal operation was reported the patient died. We presume that this operation was done along the conventional lines used for resection of rectosigmoid carcinoma in adults, that is, the abdominal stage was completed first and then the perineal stage was carried out (Table I), but no details were given.

Brief case reports of these two infants are as follows:

Case 1.—H. L., a 6-pound, 9-ounce male baby 24 hours old, was admitted to the Hospital of the University of Pennsylvania on August 19, 1944. Physical examination showed only a small dimple in the anal region and was otherwise negative. Roentgen-ray examination by the Wangensteen-Rice[6] method showed gas in the colon approximately 4.5 cm. from the anal dimple. The child was operated upon under open drop ether the same day. The skin was prepared from the clavicles to the toes and the child was placed on a sterile sheet. A perineal approach was first attempted, but the bowel could not be reached from below. The abdomen was then opened through a low left rectus incision. The lower end of the large bowel was found to be bound down tightly to the posterior abdominal wall. The lateral peritoneal folds were divided and the bowel freed. With the fingers of one hand in the perineal wound and the other hand in the pelvis through the abdominal incision, an opening was made from the pelvis to the perineum. The bowel was then guided bimanually down through the perineum and 5.0 centimeters of the distal portion of the large bowel exteriorized. The abdominal wound was then closed. The portion of bowel exteriorized was opened and a No. 24 F soft rubber catheter inserted. The bowel was then closed around the catheter. On the third postoperative day the exteriorized bowel was excised and the edges of the bowel sutured to the perineum.

After operation the baby was maintained with parenteral fluids for 48 hours. On the third postoperative day a skim milk formula was instituted. On the sixth postoperative day the baby had its first normal bowel movement.

The subsequent course of this patient was disappointing. Although the intestinal tract functioned satisfactorily, the child did not gain properly, and it was found that the blood urea nitrogen was elevated. After three months the child finally died on December 30, 1944, apparently in uremia. Autopsy revealed bilateral hydro-ureter and hydro-

TABLE I.—*The Mortality of Two-Stage Operations for Imperforate Anus (Type III of Ladd and Gross)*

Author	No. of Cases	Mortality %
Berman (2)	2	50
Quinland (3)	5	100
Fitchet (4)	10	80
Helwig (5)	3	100
Ladd and Gross*	43	72

* Ladd and Gross[2] report 1 case of a one stage abdomino-perineal repair which was unsuccessful.

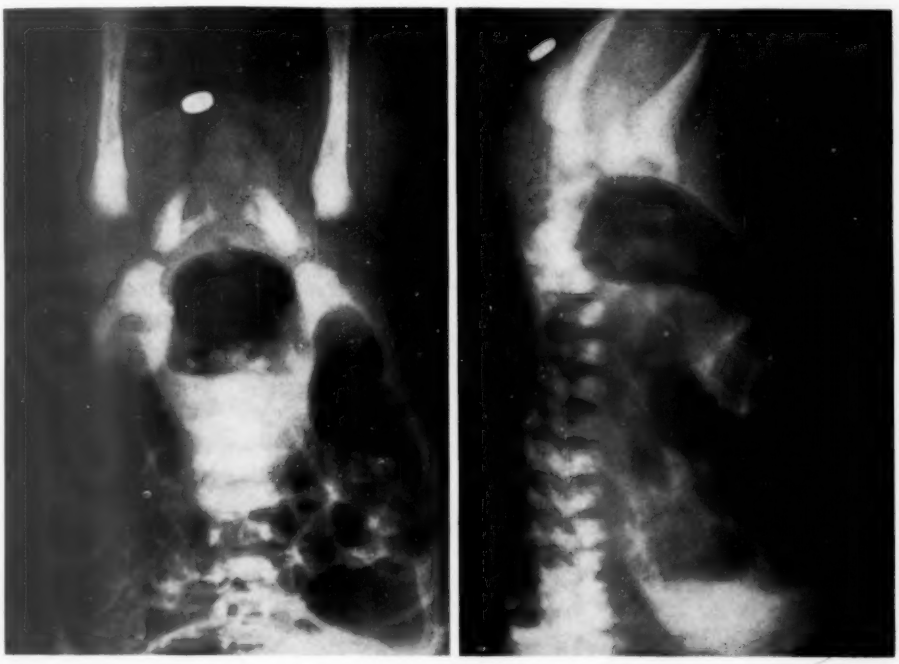


FIG. 1.—Anteroposterior and lateral roentgenograms of J. D. (Case 2) before operation obtained by the method of Wangensteen and Rice.[6]

nephrosis, but there was no evidence at autopsy that the ureters had been damaged at operation. It was thought at one time early in the patient's course that there was a urethral fistula in the perineum, but no evidence of this could be found at autopsy, and the obstruction appeared to be at the lower ends of the ureters.

Case 2.—J. D., a 5-pound, 12½-ounce male infant, was admitted to the Hospital of the University of Pennsylvania when 24 hours old with a diagnosis of imperforate anus. Physical examination revealed a small dimple where the anus should have been and a

slight hypospadias. The infant was in good condition with no evidence of dehydration. Roentgen-ray by the Wangensteen-Rice[6] method revealed a distance of approximately 7.0 centimeters between the anal dimple and the distal end of the descending colon (Fig. 1).

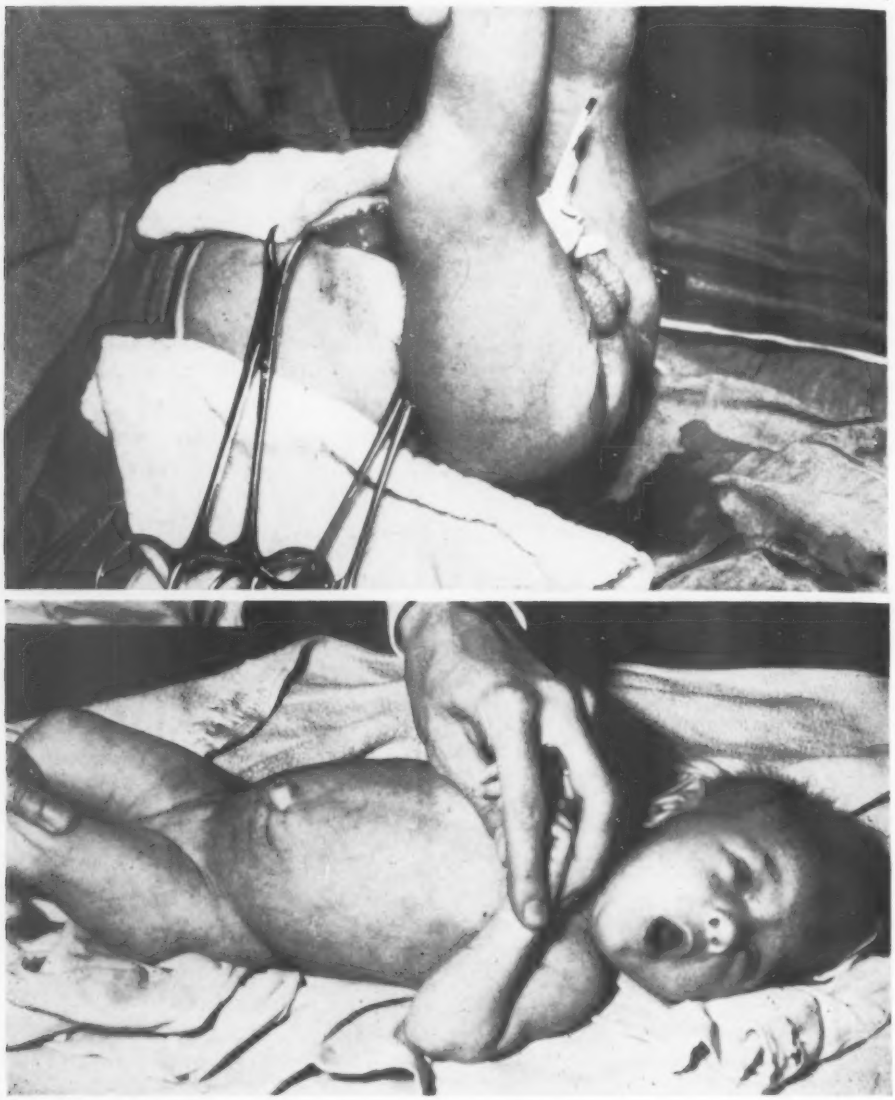


FIG. 2.—J. D. (Case 2). Position of the patient on the operating table showing both the abdominal and perineal incisions. The entire skin surface from the axillae down was prepared. The in dwelling urethral catheter is shown. Moderately distended bowel is seen in the abdominal incision.

FIG. 3.—J. D. (Case 2). The anterior wound completely healed just prior to discharge from the hospital.

A combined one-stage abdominoperineal repair as described in Case 1 was carried out, the only difference in the technic being that a small catheter was placed in the bladder to aid in preventing injury to the urethra at the time of operation (Fig. 2).

Postoperatively the infant did well, and in 36 hours was maintaining fluid balance orally. The infant remained in the hospital 29 days. At the time of discharge the infant was on an evaporated milk formula suitable for a child of this age. The perineal colostomy was functioning well. His weight at the time of discharge was 7 pounds, 0.5 ounce. Figures 2 and 3 show the wounds shortly before he left the hospital.

At two and a half months of age, the child weighed 12 pounds, 8 ounces, and the perineal colostomy continued to function well. At the age of eight months the weight was 18 pounds, and the child appeared healthy.

DISCUSSION

Lee[7] has commented on the frequency with which other abnormalities, particularly those of the urinary tract, are associated with imperforate anus.

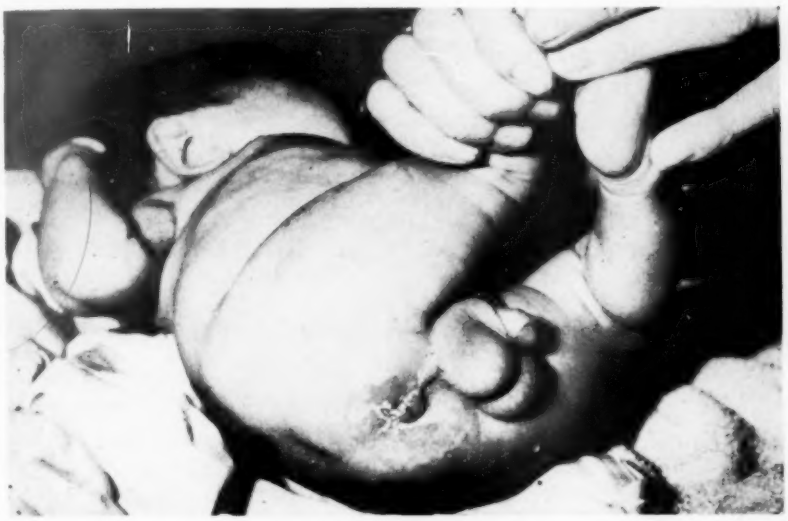


FIG. 4.—J. D. (Case 2). The posterior wound just prior to discharge from the hospital. There is still a little redundancy of the mucosa and a small amount of granulation tissue is present. The absence of sphincter control permits frequent small evacuations which have resulted in some skin irritation in spite of frequent changing of diapers. The irritation has never become severe.

As far as the pathologist could determine at autopsy, the hydro-ureters, which developed in the first case, were not the result of the operative interference but were a congenital abnormality. We felt that the survival time of three months was sufficient to indicate the safety of the operation unless there was evidence that the operative procedure was responsible for the disease of the urinary tract.

The prognosis of patients, such as the two described, will always be determined largely by the supportive treatment which they receive, and it may well be that the survival of these infants was due more to improvements in the supportive treatment than to any inherent factors in the operative procedure itself. Nevertheless, since statistics collected from the literature show that

colostomy alone has been an unsatisfactory procedure in infants with imperforate anus, we see certain advantages in the approach which was used in these two patients. First, in a certain number of the cases one would be successful in exteriorizing the bowel through the perineal wound before opening it. Second, if, through necessity or accident the bowel was opened in the peritoneal cavity, the meconium would probably still be sterile and the danger of peritonitis correspondingly less than it would be at the time of the second stage of the two-stage procedure. By doing the procedure all in one stage one does not face the possibility that the patient in whom a two-stage operation is planned will fail to improve sufficiently to permit the second stage of the operation.

CONCLUSIONS

A one-stage abdomino-perineal operation is suggested for certain cases of imperforate anus in which the colon cannot be reached safely from the perineum. The operation is facilitated by preparing the skin of the entire baby from the axillae to the toes so that the two approaches can be used simultaneously.

Two cases are reported, one child surviving three months at which time he succumbed with uremia and the other surviving and healthy at eight months of age.

The authors are indebted to Dr. A. V. Dapena for the pathological examination of H. L. (Case 1).

BIBLIOGRAPHY

1 Ladd, W. E., and R. E. Gross: Am. J. Surg., **23**: **167**: 167–183, 1934.
2 Berman, J. K.: Surg., Gynec. & Obst., **66**: 11–22, 1938.
3 Quinland, W. S.: Boston M. & S. J., **187**: 870–875, 1922.
4 Fitchet, S. M.: Boston M. & S. J., **195**: 25–31, 1926.
5 Helwig, F. C.: Am. J. Dis. Child., **38**: 551–561, 1929.
6 Wangensteen, O. H., and C. O. Rice: Ann. Surg., **92**: 77–81, 1930.
7 Lee, M. J., Jr.: Am. J. Dis. Child., **68**: 182–189, 1944.

ACUTE PANCREATITIS*

CONSTANTINE J. MACGUIRE, M.D., AND ALEXANDER J. CONTE, M.D.

NEW YORK, N. Y.

THE PRIMARY PURPOSE of this paper is to present our experiences with acute pancreatitis at St. Vincent's Hospital in the ten years between 1936 and 1946.

Acute pancreatitis is not entirely a specific diagnosis so we decided to include in our analysis only those cases which had a sudden acute clinical onset, with diagnosis confirmed either by markedly elevated blood amylase, operation or autopsy. In the cases operated upon or subjected to autopsy we included only those showing marked edema of the entire gland without rupture or a rupture of the gland with fat necrosis and bloody exudate. This cut down the number of our cases to 30, an average of three a year.

Before discussing the findings in these cases we would like to have a brief review of the subject in general.

The internal secretion of the pancreas is mainly insulin. The external secretion is (1) coagulable protein; (2) inorganic constituents; and (3) three enzymes—(a) trypsin protealytic; (b) amylase (pancreatic diastase) amylolytic; and (c) lipase (lipolytic).

We are particularly interested in amylase because in acute pancreatitis there is a temporary diffusion of amylase in the blood. Amylase is also found in small amounts in urine, lymph, feces and milk.

Seventy to 200 milligrams of sugar will normally be formed by the action of 100 cc. of blood. Values above 200 or below 60 are distinctly abnormal. Marked elevations of blood amylase from 500 units to as high as 3,000 are almost always found in the acute cases of pancreatitis, as defined previously. The peak is reached usually in 12 to 24 hours, but occasionally as late as 48 hours. After the peak there is usually a precipitous but occasionally a gradual fall to the normal level in two to six days after onset.

The absence of such an increase in serum amylase within the first six to 24 hours after the onset of acute symptoms almost certainly excludes the possibility of acute pancreatitis as the cause of the symptoms. This is not true after 48 hours. Serum lipase determinations are also very reliable but have not been so commonly used. After the initial rise the subsequent fall is more gradual than with serum amylase. In acute pancreatitis, urine values rise shortly after those in the blood. Hyperglycemia and diminished glucose tolerance occur in about 50 per cent of acute pancreatitis and glycosuria in 10 to 15 per cent.

The above brief summary of the physiologic chemistry involved in acute pancreatitis was taken from Canterow and Trumpe, Chemical Biochemistry.[1] The laboratory data in our cases is not complete so we are not able to quote percentages, only the individual findings.

* Read before the New York Surgical Society, February 5, 1947.

TABLE I

SERIES 30 CASES—ACUTE PANCREATITIS

Case No. Ad. Diag.	Sex	Age	Nat.	Predisp. Causes	Epig. Pain	Shock	Dysp.	Adm. Temp.	Nausea. Vomit	Cyan.	W & D Bl. Amyl.	P. X.	Treatment	Result
B14363 Perf. duod. ulcer	F—W 5-45	53	German	Indigestion 3 years	Severe	0	0	97.4	Nausea. Vomiting	0	28,000 80% polys. S. A. 243	Acute abd.	Expl. lap., no drain.	Disch. 11th P.O. day
31852 Acute appen.	F—W 6-47	50	Not stated	No record	Sev. post prand. 1 year	0	0	101	Nausea. Vomiting	0	14,000 81% polys. S. A. None	Acute abd.	Pancreatotomy, with drainage	Dis. 89 P.O. day—compl. thrombophl.
33639 Perf. ulcer	M—W 10-37	34	Not stated	No record	Sev. 4 hr. prior to admis.	0	0	99.4	Nausea. Vomiting	0	14,000 89% polys. S. A. None	Acute abd.	Cholocystostomy. Pancreatotomy	Disch. 26 P.O. day
22174 Perf. ulcer	M—W 7-36	42	Not stated	Chronic alcoholism	Severe	0	0	99.4	Nausea. Vomiting	0	None	Acute abd.	Pancreatotomy	Disch. 25 P.O. day
33052 Acute appen.	M—W 10-37	32	Irish	No record	General abdom. pain	0	0	101	Nausea. Vomiting	0	16,000 87% polys. S.A. None	Acute abd.	Pancreatotomy	Died at operation
32174 Perf. ulcer	F—W 9-37	59	Not stated	Vague abd. pain 2 years	Severe	0	0	99.4	Nausea. Vomiting	0	29,000 89% polys. S. A. None	Acute abd.	Cholecystostomy-Pancreatotomy	Died 7th H. D.
37745 Perf. ulcer	M—W 4-38	53	Hungarian	Indiscretion fatty foods	Severe on admis.	0	0	103	Nausea. Vomiting	0	12,400 88% polys. S. A. None	Acute abd.	Expl. lap., with drain	Disch. 27 P. O. day
36376 Acute pancr.	M—W 12-37	47	Italian	Indigestion 6 mos.	Severe epig. P.	0	0	98	Nausea. Vomiting	0	11,160 90% polys. S. A. None	Acute abd.	Pancreatotomy. Cholecystostomy	Died 66 P.O. day—ca. st. rupt.
33483 Acute pancr.	M—W 11-37	56	German	Not recorded	Severe epig. pain	0	0	100.4	Nausea. Vomiting	0	14,640 80% polys. S. A. None	Acute abd.	Supportive	Died 3rd H. D. no autopsy
7857 Acute pancr.	F—W 10-34	54	Spanish	Obese	Severe epig. P.	0	0	97	Nausea. Vomiting	0	15,700 90% polys. S. A. None	Acute abd.	Cholecystostomy	Disch. 26 P.O. day
24092 ?	M—W 6-36	31	?	Abd. cramp 6 mos. Ob. jaund.	Epig. pain	0	0	98.4	Nausea. Vomiting	0	24,600 89% polys. S. A. None	Acute abd.	Cholecystostomy. Pancreatomy	Disch. 105 P.O. day
29093 ?	M—W 6-42	44	?	Indiscretion Fatty foods	Epig. pain	+	0	98	Nausea. Vomiting	0	15,200 90% polys. S. A. 3440	Acute abd. Mass RUQ	Cholecystostomy	Died 20 P.O. day
No. ? Coroma. occlus.	M—W 25-46	64	Jewish	Obese	Epig. pain	+	+	98.6	Nausea. Vomiting	+	17,200 88% polys. S. A. 275	Abd. dist. Epig. tenderness	Miller-Abbott. I.V. fluids	Died 7 P.O.-Autop. Ac. pan.
B9689 Acute chol.	M—W 10-44	47	Italian	Obese, G. B.	Epig. pain	0	0	99.2	Nausea. Vomiting	0	16,000 85% polys. S. A. 411	Jaundice. Acute abd.	Supportive	Disch. 17—H.D.
B4337 Ac. chol.	M—W 10-44	49	Slav.	Dyspepsia for years	Epig. pain	0	0	101.6	Nausea. Vomiting	0	20,000 76% polys. S. A. None	Acute abd.	Expl. lap. 18—H.D. Fat necro.	Disch. 30 H.D.

TABLE I—(*Continued*)

TABLE I—(*Continued*)

SERIES 30 CASES—ACUTE PANCREATITIS

Case No. Ad. Diag.	Sex	Age	Nat.	Predisp. Causes	Epig. Pain	Shock	Dysp.	Adm. Temp.	Nausea. Vomit	Cyan.	W & D Bl. Amyl.	P. X.	Treatment	Result
2501 Acute append.	F—W 27–44	65	Polish	Obese	Severe on admis.	0	0	98.6	Nausea. Vomiting	0	W & D—none S. A. 207	Acute abd.	Cholecystectomy. Choledochostomy	Disch. 45 H.D.
2373 Acute append.	F—W 2–44	57	?	Obese	Severe on admis.	0	0	100.8	Nausea. Vomiting	0	29,200 86% polys. S. A. None	Acute abd.	Choledochostomy	Disch. 21 H.D.
32957 Peritonitis?	M—W 10–42	40	Irish	Known duodenal ulcer	Severe on admis.	0	0	104	Nausea. Vomiting	0	10,300 82% polys. S. A. None	Acute abd.	Drainage of pancreas	Died 2nd P.O. day
816 Perf. ulcer	M—W 12–43	46	Italian	Epig. pain 4 mos.	Severe on admis.	0	0	101	Nausea. Vomiting	0	8,000 82% polys. S. A. None	Acute abd.	Expl. lap., no drain	Disch. 17th H.D.
5169 Acute pancr.	M—W 4–44	20	Jewish	Obese	Severe	0	0	101	Nausea. Vomiting	0	10,700 73% polys. S. A. 311	Acute abd.	Cholecystectomy. Choledoc.	Disch. 39 P.O. day
16910 Perf. ulcer	M—W 8–40	75	Irish	Indigestion 1 year	General abd. pain	0	0	99.6	Nausea. Vomiting	0	14,200 80% polys. S. A. 643	Acute abd.	Expl. lap. Pancreatotomy	Died 16 H.D. aut. Ac. Pan.
A-16235 Acute chole.	F—W 10–40	69	Irish	Obese	Epig. pain	0	0	99.4	Nausea. Vomiting	0	15,250 96% polys. S. A. None	Acute epig. pain, with rebound	Expl. lap. Pancreato. Cholecysto.	Died 4th H.D. aut. Ac. Pan.
27623 Acute chole.	M—W 4–24	52	Swede	Indigestion. Obese	Epig. pain	0	0	99.8	Nausea. Vomiting	0	15,150 87% polys. S. A. 795	Acute epig. pain	Supportive	Disch. 19th H.D.
A-39085 Acute chole.	M—W 5–43	55	Irish	Indigestion. Obese	Epig. pain	0	0	100.4	Nausea. Vomiting	0	19,650 80% polys. S. A. None	Acute abd.	Expl. lap., with drain	Died 28 H.D. autop. Ac. Pan.
17024 Acute append.	M—W 11–40	39	Irish	Chronic alcoholic	Epig. pain	0	0	102.4	Nausea. Vomiting	0	10,500 82% polys. S. A. None	Acute abd.	Expl. lap., with drain	Died 3rd H.D. autop. Ac. Pan.
16330 Twisted ovarian cyst	F—W 9–40	41	Italian	Epig. pain 2 mos. Obese	Epig. pain	Acute	+	98.6	Nausea. Vomiting	0	40,200 73% polys. S. A. 900	Acute abd.	Supportive. Bl. trans.	Disch. 24 H.D.
16898 Cerebral acc.	F—W 1–43	47	Italian	Obese. Jaundice 2 wks. and rex.	0	Coma	+	97	Nausea	+	0	General abd. pain	Supportive	Died 1st H.D. AP
44363 Gastro-enter.	M—W 12–43	32	?	Chronic alcoholic	Epig. pain	0	0	99.6	Nausea. Vomiting	0	14,700 88% polys. S. A. 474U	Acute abd.	Supportive	Died 1st H.D.
29722 Intest. ob.	M—W 7–42	33	Polish	Constipation. Abd. disten.	Epig. pain	0	0	98.6	Nausea. Vomiting	0	4,600 52% polys. S. A. 200U	Acute abd.	Expl. lap., with drain	Transfer

TABLE II

SPECIAL CASE

Case No. Adm. Diag.	Sex	Age	Nat.	Predisp. Cause	Epig. Pain	Shock	Dysp.	Adm. Temp.	Nausea. Vomit	Cyan.	W & D Bl. Amyl.	P. X.	Treatment	Result
29875	M 6–42	39	Chinese	Indiscretion. Fatty foods. RUQ dist.	None	0	0	99	Nausea. Vomiting	0	None	Acute abdomen	Cholecystectomy (empyema)	Disch. 33rd H.D.
Re-ad.	8–42				Acute epig. pain	Acute shock	+	97.6	Nausea. Vomiting	0	None S. A. 836, 854	Board-like abdomen	Exp. lap. Acute hemorrhagic pain, with fat necrosis	Disch. 16 H.D.
Re-ad.	7–43				Acute epig. pain—radiates to back	0	0	98	Nausea. Vomiting	0	S. A. 377	Acute abdomen	Supportive	Disch. 5th H.D.
Re-ad.	9–43				Epig. pain	0	0	100	Nausea. Vomiting	0	S. A. 663	Acute abdomen	Supportive	Disch. 9th H.D.
Re-ad.	10–43				Epig. pain	0	0	99.6	Nausea. Vomiting	0	S. A. 522	Acute abdomen	Supportive	Disch. 7th H.D.
Re-ad.	7-28-44				Epig. pain	0	0	100	Nausea. Vomiting	0	S. A. 900 15,200 78% polys.	Acute abdomen	Supportive	Disch. 6th H.D.

ACUTE PANCREATITIS

ETIOLOGY

Fitts, of Boston, in 1889, published the first clear-cut pathologic and clinical description of acute hemorrhagic pancreatitis. The etiology, however, is still undetermined. Trauma, vascular injury, infection and bile invasion of the pancreatic ducts are all factors.

About two years ago John Morton,[2] in his paper published in Surgery, had a very complete bibliography on this whole subject, with particular references to Somogyi's[3] contribution to diagnosis in 1938, when the simple method of amylase determination was developed. In connection with pathology, however, he placed great importance on the differentiation between acute edematous pancreatitis and acute necrosis, a differentiation which affects treatment. Where

TABLE III

MORTALITY PERCENTAGE—NO OPERATIONS

Total Cases..... 7 Recovery..... 3 Died..... 4

Choledochostomy	Expl. Lap., with Drain.	Pancreatotomy	Cholecystostomy, with Pancreatotomy	Cholecystectomy, with Choledochostomy	Cholecystectomy
Cases..... 1	Cases..... 7	Cases..... 5	Cases..... 5	Cases..... 2	Cases..... 3
Recovered.. 1	Recovered.. 5	Recovered.. 4	Recovered.. 2	Recovered. 2	Recovered. 3
Died..... 0	Died..... 2	Died..... 1	Died..... 3	Died..... 0	Died..... 0

Total Operated Cases	23
Recoveries	17
Deaths	6
Total Females	9
Total Males	21
Average Age	47.5
P. H. Gallbladder disease	15
P. H. Obesity	11
P. H. Alcoholism	3

the diagnosis of edema without necrosis was made 29 cases not operated upon all recovered. In 27 cases where pancreatic necrosis was diagnosed or discovered, 19 died—ten with operation and nine without operation. He does not, however, make clear how the diagnosis between edema and necrosis is made except by the fact that the cases of edema improved and the cases with necrosis did not. He feels the cases with necrosis should be operated upon for drainage if nothing else. The cases of necrosis are more prostrating at the start, but this is a matter of degree of symptoms.

All of us who have been on call for emergency surgery in an acute hospital have tried to keep in mind the diagnosis of acute pancreatitis in any sudden abdominal crisis, and still, strangely enough, the diagnosis was rarely made before operation until the advent of amylase determination. The clinical picture varies, and we cannot do better than quote three personal experiences of T. M. Furber,[4] of Sydney, Australia, as follows:

"I well remember a man at Sydney Hospital who, while waiting in Dr. Hamilton Marshall's out-patient department, fell from his seat, laboring under what Lord Moynihan has so aptly called, 'illimitable agony.' It was evident even to such mole-eyed students as we were that the unfortunate old man was stricken unto death, as he quickly became a dreadful, livid, ashen hue, and writhed and groaned in a calamity of pain, fighting for

breath and calling on our Maker for help in what was truly his last extremity. Some three days later he died, as was discovered at autopsy, of pancreatic necrosis.

"Another picture which comes to mind is that of a decent, middle-aged widow, who lived virtuously alone, with gallstones, which for years, in spite of advice, she had harbored and, I think, cherished. Soon after dinner one night she was overwhelmed by an abdominal cataclysm. When I saw her a few hours later she was sitting up in the middle of a big double bed, slowly rocking herself back and forth and gently moaning, but not taking the slightest notice of the bewildered females who clucked and fluttered around her, nor answering direct questions, for her sensorium was apparently saturated by the

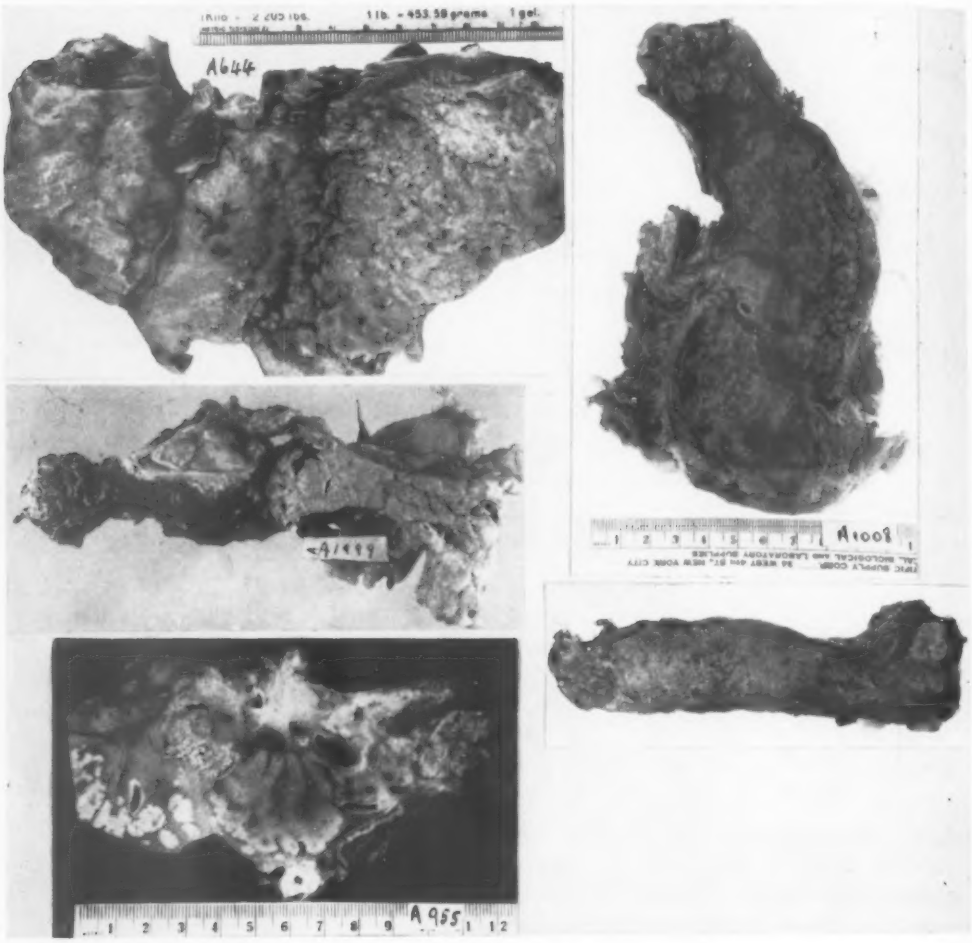


FIG. 1.—Photographs of five gross specimens of Acute pancreatitis removed at autopsy.

tremendous discharge of afferent stimuli from her abdomen. Knowing that she had gallstones, I guessed that she had acute pancreatitis, and advised her immediate removal to the hospital; but while arrangements were being made her tempestuous daughter burst in on us like a southerly gale and whisked the unfortunate old lady to her own home, where the poor soul perished of what was proved, at the coroner's autopsy, to be pancreatic necrosis.

"Fortune seemed to smile on a sprightly, brisk, young bachelor when he won the lottery. He at once gave up his uninteresting and comparatively unremunerative toil for what was, for him, the more congenial task of helping the brewery proprietors to issue a few more bonus shares. He succeeded so well in this, no doubt, laudable crusade that for a month or more he was seldom sober, and thought he was having a wonderful time, till

suddenly the Grim Reaper breathed on his pancreas and it died, and so did its owner within three days. He, unlike the other two, lay as still as a mouse or as a patient with a ruptured peptic ulcer. The metabolic revolution had caused such profound shock that, as Professor Wilson used to say: 'It was patent to the meanest intelligence' that the poor fellow was destined soon to find out whether the story of Saint Peter and the Digger had any truth in it. Resuscitatory treatment did not reclaim him, and he died without operation. Autopsy revealed hemorrhagic pancreatitis."

It is difficult to add to this complete and dramatic clinical description, but there are three features which have impressed me in differentiating this condition from a perforated ulcer: (1) Shock, which is usually absent in ulcer. (2) Rigidity more or less confined to the upper abdomen and not diffuse as in ulcer. (3) Occasional cyanosis.

Tables I and II represent the summary of cases from the surgical service at St. Vincent's Hospital, selected as explained in the opening paragraph. (Table II is on a special case.) In Table III certain features deserve emphasis: The predominance of males over females—21 to 9—is just reverse of the findings in other series; also, strangely enough, the operative cases had a lower mortality than the nonoperative cases. Pancreatotomy, which is distinctly contraindicated, had a mortality of only 20 per cent. Fifteen cases gave a previous history of gallbladder disease. This seems to be in line with previous reports. To have significance, however, the mortality statistics should be broken down into pathologic groups. This we have not been able to do in this series. The five photographs were taken at autopsies on cases included in this review, and were taken by our pathologist, Dr. Rottino.

Many gaps in the clinical data are obvious in this statistical review. It is too bad we can not plan these analyses ahead rather than doing them in retrospect. Under conservative treatment abscesses not infrequently form at the end of a week or ten days, which, upon drainage, discharge large amounts of foul-smelling material, somewhat like rancid curds of milk.

This sequela has a very definite mortality of its own. When the diagnosis of acute pancreatitis is made operation should be deferred, but it is a question whether delayed operation seriously affects the mortality. I am convinced that in some cases cholecystostomy and drainage of the lesser sac are distinctly beneficial. Pancreatotomy should never be undertaken, it provides no real drainage and may lead to disastrous hemorrhage.

CONCLUSIONS

1. Amylase readings are essential to the diagnosis of acute pancreatitis.
2. The edematous type should not be operated upon.
3. The necrotic type should be operated upon after the initial shock subsides.
4. The clinical distinction between the two types is not easily made.

REFERENCES

[1] Canterow and Trumpe: Chemical Biochemistry.
[2] Morton, John: Acute Pancreatitis, Surgery, 17, No. 4, April, 1945.
[3] Somogy, M.: Micromethods for Estimation of Diastase. J. Biol. Chem., 125, 399-414, 1938.
[4] Furber, T. M.: Medical Journal of Australia, p. 405, October 26, 1940.

BURSITIS UNDER FIBULAR COLLATERAL LIGAMENT

ANTHONY F. DE PALMA, M.D.
PHILADELPHIA, PA.

THE PURPOSE OF THIS PAPER is to present six cases of chronically inflamed bursae found under the fibular collateral ligament of the knee joint. Voshell and Brantigan demonstrated such a lesion in the region of the tibial collateral ligament and Hendryson recently reported six cases of bursitis of the fibular collateral ligament. Cystic masses at the level of the knee joint line along the outer aspect are prone to be associated with cystic degeneration of the external meniscus. However, careful dissection of these lesions and microscopic studies reveal that some of these masses are in no way related to the meniscus but are nothing more than chronically inflamed bursae. Piersol describes two bursae in this region, one found between the fibular collateral ligament and the tendon of the popliteus muscle; and the second, lies between the fibular collateral ligament and the tendon of the biceps femoris. He further describes the possibility of a bursa at the point where the tendon of the popliteus muscle merges at its insertion with the capsule of the knee joint. It is at this point that these painful masses are located, and one may readily conclude that the painful cystic masses described by Hendryson and the six reported in this paper may be traumatized hypertrophic bursae which are normally found in this area.

CLINICAL PICTURE

The lesion is usually found in the male sex. Age is of no significance for in this small group the age range was from 16 years to 42 years. Usually there is a history of trauma; however, in one case reported no such history could be obtained. Repeated mild traumata undoubtedly is a definite causative factor. Pain and the presence of a mass on the outer aspect of the joint are constant findings. The pain is aggravated by walking, standing, and exercising. In some instances the pain is more severe at night. The mass, always present, may appear smaller at one time and larger at another. The patient also has a feeling of "tightness" within the knee joint. In no case was there diffuse swelling of the joint or a history of locking or instability of the knee joint. The duration of the symptoms was from six months to three years.

Physical examination usually reveals a small, tense, round, cystic mass from 1½ cm. to 3 cm. on the anterior aspect of the fibular collateral ligament at the level of the knee joint line. The mass seems to dip beneath the anterior border of the ligament. The mass fails to disappear upon pressure with the knee flexed. It becomes more tense upon extension of the joint and upon hyperextension pain is greatly intensified. Pain and tenderness is elicited by direct pressure over the lesion and upon adduction of the tibia on the femur.

Occasionally there may not be a distinct mass present, as in Case II. In this instance no circumscribed mass was present but the fibular ligament

appeared to be distended at the level of the joint space and palpation of this area revealed a tumefaction of the underlying tissues. Pressure elicited tenderness, and hyperextension produced exquisite pain. The tumefaction failed to disappear upon flexion of the knee joint.

CASE REPORTS

Case 1.—E. H., male, age 42. The patient had pain in the outer aspect of the right knee for six months. There was no history of trauma. The pain had an insidious onset and was becoming progressively more severe. He was conscious of a feeling of "tightness" on the outer aspect of the knee joint. Within the last month the pain was worse at night and aggravated by standing and walking. He gave no history of locking or instability of the joint.

Physical examination revealed a circumscribed mass 1½ by 3 cm. anterior to the fibular collateral ligament at the joint line. One-third of the mass dipped beneath the anterior margin of the ligament. The mass failed to disappear or decrease in size upon flexion of the knee joint; upon extension the mass became more tense and extension and hyper-extension of the joint elicited pain; adduction of the tibia, as well as direct pressure over the lesion, also produced severe pain. Roentgen-ray examination was negative for evidence of bone pathology. A diagnosis of bursitis was made.

At operation a mass 1½ by 3 cm. was found lying anterior to, and under, the anterior margin of the fibular collateral ligament. It was dissected out in toto without opening the joint cavity. There was no connection between the mass and the external meniscus. It was, however, loosely attached to the capsule of the joint. The patient made an uneventful recovery and was completely relieved of all symptoms.

Case 2.—P. P., male, age 17. The patient had a painful swelling on the outer aspect of the left knee for eight months. He gave a history of repeated traumata to the left knee joint during the previous football season. The pain was aggravated by walking and standing. He had no history of locking or instability of the joint.

Physical examination revealed a localized swelling on the outer aspect of the left knee joint. The swelling could not be defined by palpation, but the fibular collateral ligament was taut. The tumefaction did not disappear upon flexion of the knee joint. The pain was aggravated by extension and hyperextension of the joint, and by adduction of the tibia. Pressure over the area elicited marked tenderness. Roentgen-ray examination was negative for evidence of bone pathology. A diagnosis of bursitis was made.

At operation the mass was exposed by a longitudinal incision through the fibers of the fibular collateral ligament. Upon division of the fibers the mass herniated into the wound. The posterior surface was firmly adherent to the capsule of the knee joint. During the dissection the external meniscus was exposed. Inspection of the meniscus revealed no pathologic changes in this structure. The patient made an uneventful recovery and played football the following season.

Case 3.—W. Q., colored male, age 42. Three years prior to admission the patient was struck on the outer aspect of the left knee by a falling box weighing about 60 pounds. The knee became painful and swollen. He was treated by rest and a compression bandage. He noticed, after all swelling had subsided, a small mass on the outer aspect of the left knee joint which was painful on pressure and caused considerable discomfort at night. A diagnosis of a "cyst" was made and two attempts were made to remove the mass surgically. The mass reappeared after each surgical procedure. He was admitted to the Jefferson Hospital six months after the last operation complaining of a painful knee.

Physical examination revealed a large nodular mass on the outer aspect of the left knee. The mass seemed to disappear under the fibular collateral ligament. It was fixed to the underlying tissues and had a doughy feeling. Pressure over the mass elicited

tenderness. It failed to disappear upon flexion of the joint. Extension and hyperextension made the mass more tense and more painful. Roentgen-ray examination was essentially negative except for a soft tissue shadow cast by the mass outside the joint.

At operation the mass was exposed by a longitudinal incision. It was found immediately below the skin and appeared to dip beneath the lateral ligament. The part of the mass anterior to the ligament was readily delivered, but the posterior one third of the mass which lay beneath the ligament was delivered with great difficulty. It was necessary to divide some of the anterior fibers of the ligament in order to obtain an adequate exposure. The specimen measured 5 by 4½ cm. and was external to the joint capsule. The meniscus was not exposed.

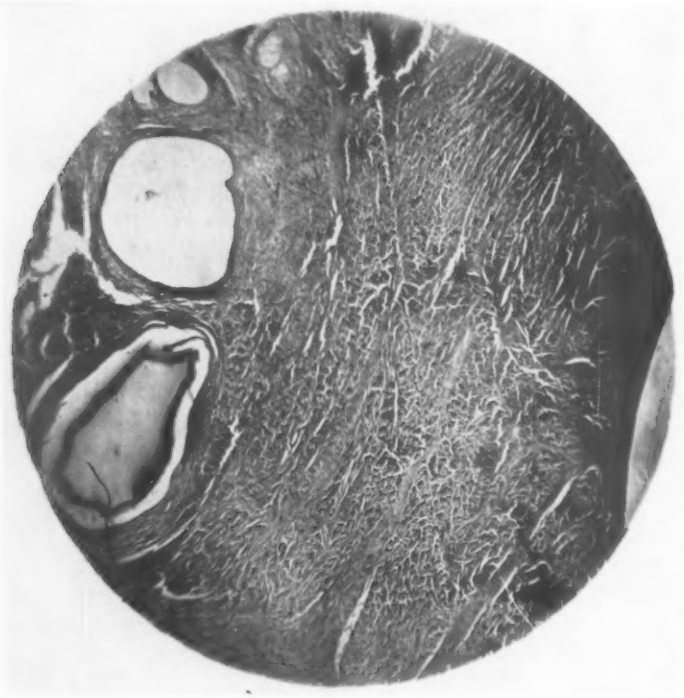


FIG. 1.—The microscopic section is typical of the histo-pathologic changes found in all six cases presented. Note the marked increase in fibrous tissue, and throughout there are many small cyst-like areas. These cysts have thickened fibrous tissue walls and are lined by a layer of flattened endothelial cells. The findings are consistent with chronic inflammation of a bursa.

Case 4.—N. B., white, male, age 26. This 26-year-old veteran first injured his left knee two years ago when he struck the joint against the side of a jeep while getting into the vehicle. The joint became somewhat swollen and painful, but not enough to require hospitalization. About six months later he noticed a small mass on the outer aspect of the joint which was rather painful, more so at night. About that time he fell on the knee during maneuvers and this time his knee became so painful that he was hospitalized for three weeks before he was returned to duty. The painful mass persisted and within the past month he has been unable to walk without severe pain.

There was a mass 2½ by 3 cm. on the outer aspect of the left knee joint anterior to the fibular collateral ligament. Pressure over the mass elicited tenderness. The mass did not disappear on flexion of the joint. Extension and hyperextension aggravated the

pain and made the mass more tense. No limitation of motion of the joint was present, but both active and passive motion produced an audible "thump."

At operation a nodular mass was found under the skin which extended beneath the fibular ligament. It was so firmly adherent to the capsule that the capsule was removed with the mass and the periphery of the cartilage was exposed. The joint was explored and a discoid cartilage was found which was removed. Macroscopically no evidence of cystic degeneration was found in the cartilage. Microscopic sections of the meniscus also failed to reveal any cystic degenerative changes.

Case 5.—A. C., white, male, age 28. This patient, an interne at the Jefferson Hospital, sustained several direct injuries to the outer aspect of the left knee while playing football two years prior to his present admission. He again injured the same knee during maneuvers while in the service. A diagnosis of a "cyst" was made and a mass was removed in an army hospital. He was free of symptoms until six months before admission to the hospital, when he again noticed a small mass on the outer aspect of the left knee. The mass became progressively larger and was very painful if inadvertently struck. His pain was constant, more severe at night; it was aggravated by standing and walking. The mass was aspirated on two occasions and a few cubic centimeters of a yellowish serous fluid were obtained each time. Following the aspiration some relief resulted and the mass diminished in size. However, it became distended and painful again within a few days.

Physical examination showed a round distended mass 2 by 2½ cm. on the lateral aspect of the left knee immediately under a three-inch vertical scar anterior to the fibular collateral ligament. The scar was firmly adherent to the underlying mass which was nodular and firmly attached to the underlying tissues. The posterior margin was not palpable, for it appeared to dip under the fibular ligament. Motion of the knee was not restricted. Extension and hyperextension of the joint made the mass more tense. Uniform pressure over the mass failed to reduce its size nor did it become smaller or disappear upon flexion of the joint.

At operation the old scar was resected and the mass was found so firmly attached to the capsule that a portion of the capsule was removed during the dissection, and the periphery of the meniscus was exposed. The cartilage appeared normal. No evidence of any cystic degeneration was found. In order to completely remove the tumor a second incision was necessary paralleling the posterior border of the fibular ligament. The mass was then readily delivered in toto from underneath the fibular collateral ligament.

Case 6.—J. H., white, male, age 17. During the last high school football season the patient sustained repeated injuries to the left knee. Toward the end of the season he noted a small painful lump on the outer aspect of the joint. The mass became progressively larger and more painful. He was treated by a compression bandage. The mass became smaller but did not disappear. Within the past two months the knee had become very painful, especially when walking. The pain was worse at night. He was unable to find a comfortable position for his leg.

Physical examination revealed a small, rounded, smooth mass 1½ by 2 cm. on the outer aspect of the left knee joint anterior to the fibular collateral ligament. The mass was firm, lay immediately beneath the skin, and was firmly attached to the underlying tissues. Pressure elicited pain. The mass did not disappear when firm pressure was made upon it. Hyperextension of the joint increased the pain and made the mass more prominent.

At operation the mass was exposed by a small two-inch vertical incision and peeled off the capsule of the joint without exposing the meniscus. Its posterior border was found under the fibular ligament but it shelled out very readily after the ligament was retracted posteriorly. Section of the mass revealed that it was composed of several loculated areas which contained a thick colorless, jelly like substance.

REMARKS

It is of interest to note that in one case (Case 4) a discoid cartilage was present. Besides the painful mass present on the lateral aspect of the knee, a a definite audible "thump" could be heard on active and passive flexion and extension of the knee. The cartilage could readily be felt, with each excursion of motion, to protrude beyond the articular margin of the outer tibial plateau. The forward displacement of the cartilage caused definite compression of the bursal sac, for the mass became more tense and the tibular fibular ligament more taut.

It is obvious, in this instance, that the hyper-mobile discoid cartilage was the exciting factor in the formation of the cystic mass. The repeated traumata to the bursal sac resulted in a hyperplasic, thickened, loculated, chronically inflamed bursa.

Complete removal of the entire mass is essential in order to bring about a cure. In Cases 2 and 5 the mass recurred, following excision. Case 2 had had two previous excisions. Case 5 had had one previous excision. Frequently the bursa lies deep beneath the tibular fibular ligament and part of it may not be accessible because of the difficulty encountered in retracting the taut ligament sufficiently to give adequate exposure of the lesion. It may be necessary in such instances to split the tibular fibular ligament longitudinally in order to obtain access to the entire mass. On the other hand, it may be necessary to make a second incision parallel with the posterior border of the tibular fibular ligament and retract the ligament anteriorly in order completely to excise the mass.

REFERENCES

Voshell, A. F., and O. C. Brantigan: Bursitis in the Region of the Tibial Collateral Ligament. J. Bone and Joint Surg., **26**: 793–798, 1944.

Hendryson, I. E.: Bursitis in Region of Fibular Collateral Ligament. J. Bone and Joint Surg., **28**: 446–450, 1946.

The Editors and the Publishers of the ANNALS OF SURGERY regret to announce the death of Dr. Roscoe R. Graham of Toronto, Canada on January 17, 1948. Doctor Graham served as a member of the Editorial Board for many years. His death will prove a great loss to the ANNALS OF SURGERY. A suitable memoir will appear in a later issue of the ANNALS.

TRAUMATIC ANEURYSM OF THE SUBSCAPULAR ARTERY*

ROBERT A. WISE, M.D.**
PORTLAND, OREGON

FROM THE SURGICAL SERVICE OF THE VETERANS HOSPITAL, PORTLAND, OREGON

THE OCCURRENCE OF AN UNUSUAL arterial lesion among the large group of patients sustaining vascular injuries in World War II is not difficult to understand. Modern shells, which broke into innumerable small fragments travelling at high velocity, produced such an unprecedented number of vascular injuries that among them uncommon lesions can be expected. The only reports of subscapular aneurysm found in the literature are by Liston[1] in 1820 and McGraw[2] in 1865. Matas[3] encountered none in his large series of aneurysms, and Makins'[4] collection of vascular injuries in World War I listed only two of the subscapular artery. Elkin[5] has not included one in his publications from the Army Vascular Center. The rarity of the lesion is due to the anatomic location of the artery. The subscapular artery is a branch of the third portion of the axillary artery and lies on the subscapularis muscle in close association with the chest wall medially. A perforation of the artery in the distal two-thirds of its course would probably close spontaneously because the small fascial space would limit expansion of a blood clot, while in the proximal third, near its origin, an associated perforation of the axillary artery would be likely. An aneurysm of the subscapular artery can be expected to occur only when the artery is injured near its origin, without associated injury to the axillary vessels. The following case report describes such an occurrence.

CASE REPORT

C. N., white, male, age 33, entered the Portland, Oregon Veterans Hospital on September 2, 1946. He had been hit in the right arm by a shell fragment in February, 1945, while in Germany. The wound of entrance was on the lateral surface of the arm in the mid-deltoid region. There was no wound of exit, the shell fragment lodging in the chest wall high in the axilla. There was considerable hemorrhage from the wound, which stopped with pressure. However, the entire right axilla and shoulder became markedly swollen and ecchymosis appeared, extending to the elbow and onto the chest wall posteriorly in the scapular region and anteriorly to the sternum. The patient was evacuated to a general hospital in England where two massive hemorrhages from the external wound occurred on the 10th and 12th days following the initial injury. There was no further hemorrhage, the massive swelling of the shoulder decreased and the external wound closed. However, the patient noted a small circumscribed, firm swelling in the right axilla, which persisted and was present when he returned to civilian life. This axillary swelling gradually increased in size and became markedly enlarged during heavy work. It was associated with axillary pain, which radiated along the inner surface

* Published with permission of the Chief Medical Director, Department of Medicine and Surgery, Veterans Administration, who assumes no responsibility for the opinions expressed or conclusions drawn by the author.

** Formerly Colonel, M. C., A. U. S.; Chief of Surgery, 203rd Gen. Hosp. France.

of the right arm and forearm to the wrist. The skin over the mass became red, tender and inflamed.

Physical Examination.—Examination on admission revealed a tense, compressible, pulsating swelling in the right axilla, measuring 8 x 5 x 6 cm. There was no fremitus. Auscultation over the mass disclosed a soft, systolic bruit which disappeared on compression of the right subclavian artery. The right and left radial pulses were equal in volume and no dilatation of the veins of the right arm or forearm was present. The blood pressure was equal in both upper extremities: systolic 132, diastolic 80. Obliteration of the aneurysm caused no elevation in blood pressure or slowing of the pulse rate. A teleoroentgenogram of the heart showed no enlargement.

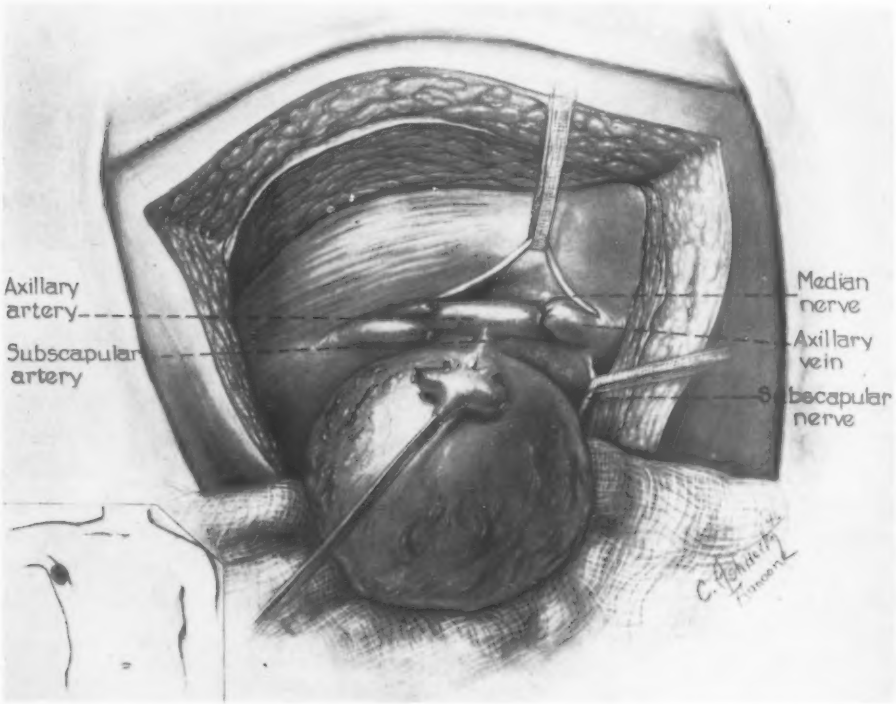


FIG. 1.—Findings at operation. The subscapular artery communicated with the sac through two openings.

Operation.—September 5, 1946. Under nitrous oxide oxygen ether anesthesia, an incision was made from the outer third of the clavicle across the anterior axillary fold onto the upper third of the arm. The pectoralis major and minor were cut near their insertions, exposing a large, pulsating mass, closely associated with the axillary artery and vein and occupying the entire subscapular region of the axilla. The origin of the mass could not be determined accurately at this point, but it appeared to arise from the axillary artery. Controlling tapes were placed around the axillary artery and vein, proximal and distal to the mass, but were not tied. Dissection of the aneurysm was commenced at its lowest extent on the subscapularis muscle and continued upward to the axillary vessels. The subscapular nerve, densely adherent to the sac, was isolated and protected. At the apex of the mass, the axillary vein was so incorporated into the wall of the sac that it could not safely be dissected free. Therefore, after ligation, a portion of the axillary vein was retracted with the sac. The origin of the aneurysm was now apparent (Fig. 1). It arose from the subscapular artery 1.5 cm. from the axillary artery.

After complete dissection, the aneurysm now hung from the subscapular artery similar to a pear on a bough. The axillary artery represented the branch, the subscapular artery the stem, and the aneurysmal sac the fruit. It was possible to ligate and divide the subscapular artery between the sac and the axillary artery, making removal of the aneurysm possible without interrupting the continuity of the axillary artery.

The specimen consisted of an aneurysmal sac, measuring 8.5 x 6 x 5 cm. to which a segment of the axillary vein was attached. The subscapular artery was adherent to the apex of the sac and continued along its posterior wall. It communicated with the sac through two openings 1 mm. in diameter. There was no communication between the sac and the axillary vein. The inner lining of the aneurysm was smooth and its lumen was partially filled with laminated blood clot.

Postoperative Course.—The postoperative course was smooth and the patient was discharged on the 14th postoperative day.

Follow Up.—February 8, 1947. The axillary incision was firmly healed and there was complete range of motion of the right shoulder. There was no swelling of the right arm or forearm and the brachial and radial pulses were present. There was no recurrence of the aneurysm. The patient had returned to work as a logger.

DISCUSSION

Recently emphasis has been placed on the importance of maintaining the continuity of the main arterial channel, whether by endoaneurysmorrhaphy, lateral suture, end to end anastomosis or vein graft, in the surgical treatment of traumatic vascular lesions. Even should the collateral circulation be adequate for the viability of the peripheral tissues, complete division of the major vessel often results in lowered work capacity and intermittent claudication. It is not difficult to maintain the integrity of the major vessel when a vascular lesion arises from one of its branches. Ligation of the branch with excision of the aneurysm can be accomplished without disturbing the main vascular channel. However, to make this possible, established technical procedures of vascular surgery must be observed: absolute hemostasis, wide exposure, proximal and distal control of the main vessel, and meticulous dissection are imperative.

SUMMARY

1. A case of traumatic aneurysm of the subscapular artery is reported.

2. Ligation of the subscapular artery and excision of the aneurysm, without interrupting the continuity of the axillary artery, were possible.

BIBLIOGRAPHY

[1] Liston, R.: Ossified Aneurysmal Tumour of the Subscapular Artery. Edinburgh Med. & Surg. J., **16**: 66, 215, 1820.

[2] McGraw, T. A.: A Case of Traumatic Aneurysm of the Subscapular Artery. New York Med. J., **41**: 560, 1885.

[3] Matas, Rudolph: Personal Experiences in Vascular Surgery, A Statistical Synopsis. ANNALS OF SURGERY, **119**: 802, 1940.

[4] Makins, George H.: Gunshot Injuries to the Blood Vessels. Bristol, John Wright & Sons, Ltd., 1919.

[5] Elkin, D. C.: Vascular Injuries in Warfare. ANNALS OF SURGERY, **120**: 284, 1944.

[6] Bigger, I. A.: Treatment of Traumatic Aneurysms and Arteriovenous Fistulae. Arch. Surg., **49**: 170, 1944.

[7] Freeman, N. E.: Arterial Repair in the Treatment of Aneurysms and Arteriovenous Fistulae. ANNALS OF SURGERY, **124**: 288, 1946.

CYST OF THE SPLEEN

Case Report

V. F. Lang, M.D., S. A. Morton, M.D., J. D. Steele, M.D., and A. A. Schaefer, M.D.

Milwaukee, Wis.

Primary cysts of the spleen, as distinguished from those occurring as a result of degenerative processes due to disease or trauma, are apparently rare. In the case herein reported, the presenting symptoms and signs were of thoracic origin. As far as we have been able to determine, this is the only case in which the spleen containing a cyst has been successfully removed through a transthoracic approach.

Mrs. M. M., 39 years of age, at the time of our initial observation on April 13, 1946, was six weeks pregnant, this being her first pregnancy. She was seen first because of pain in her chest. Physical examination of the chest, at this time, was negative. Five days later, a roentgenogram of her chest, taken because of pain and bronchial breathing in the left chest, showed considerable density at the left base. Evidence of effusion subsequently appeared and a thoracentesis yielded clear fluid which was negative on culture and guinea pig inoculation for mycobacterium tuberculosis.

She was admitted to Columbia Hospital on April 29, 1946. A roentgenogram of her chest, taken the morning after admission, showed the same appearance at the left base as previously. Some hours later, she complained of sudden, sharp pain in the left chest with respiratory distress. A marked increase in the effusion was noted on roentgenologic examination. Thoracentesis now yielded rather heavy, turbid fluid which was sterile on culture. The fluid contained many cholesterol crystals but no cells. This latter finding suggested that the fluid might be of a cystic origin (Fig. 1). For the next few days, there was no increase in the pleural effusion, but there was an increase in her fever and vaginal bleeding appeared. Since it was evident that the patient was aborting, a curettage was accordingly performed on May 3, 1946, five days after admission to the hospital.

As her symptoms improved, more adequate roentgenologic examination was possible. In addition to the effusion in the posterolateral aspect of the left chest, a spherical, calcified lesion was seen in the region of the left side of the diaphragm, the exact position of which in relation to the diaphragm could not be accurately determined, at this time (Figs. 2 and 3). Subsequently, the stomach was outlined with barium and a gas producing substance was given by mouth to determine the relation of the stomach to the calcified lesion. A diagnostic pneumoperitoneum was also induced. The preoperative roentgenologic diagnosis was a cystic mass with a calcified wall in the abdomen on the left side just below the diaphragm. The contents of the cyst were believed to have erupted through the diaphragm and to have secondarily involved the pleural cavity.

On May 20, 1946, three weeks after admission to the hospital, a thoracotomy was performed under pentothal anesthesia, through a posterolateral incision. The 9th rib was resected from the transverse process to the anterior axillary line. When the pleura was opened, a large amount of turbid fluid containing fibrin was encountered. The visceral pleura was moderately thickened. The diaphragm was incised and a large cyst encountered which contained thick, yellowish, granular material. Since it was believed that the cyst arose from the spleen, splenectomy was performed. Air tight, intercostal drainage was instituted after closure of the diaphragm and chest wall.

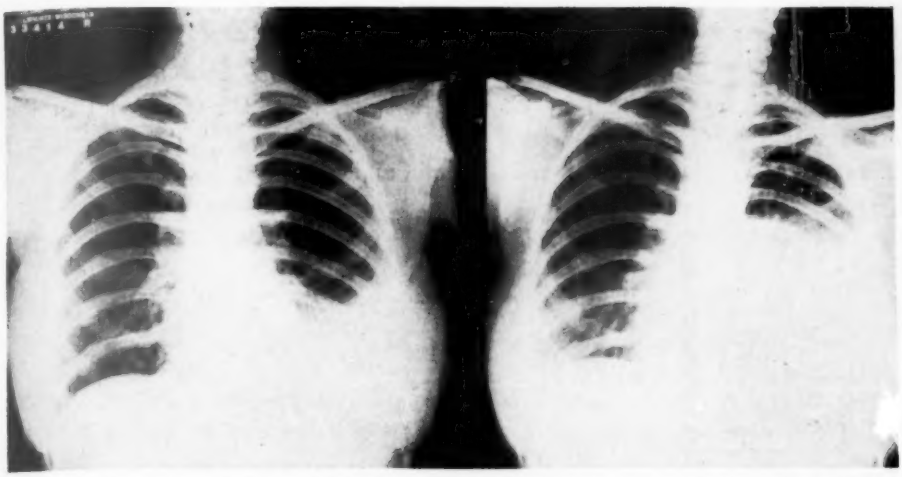


FIG. 1.—Plates taken before and after acute episode. Note increasing fluid at the left base.

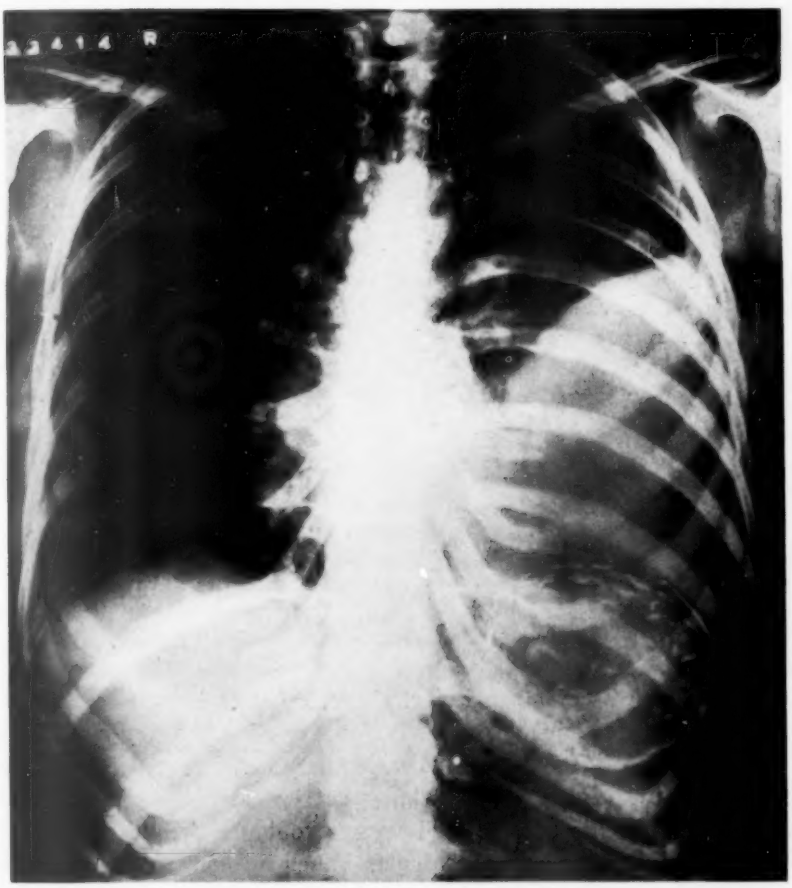


FIG. 2.—Bucky diaphragm plate showing calcified cyst, position indeterminate.

The patient's immediate postoperative course was uneventful except for a high fluctuating fever which continued for five days. She was discharged from the hospital 18 days following the operation, in good condition.

Pathologic examination by G. H. Hansmann, M. D., revealed that the gross specimen (Fig. 4) consisted of a spleen which weighed 305 Gm. and measured 15 cm. in length, 10 cm. in width, and 3 cm. in thickness. The lower

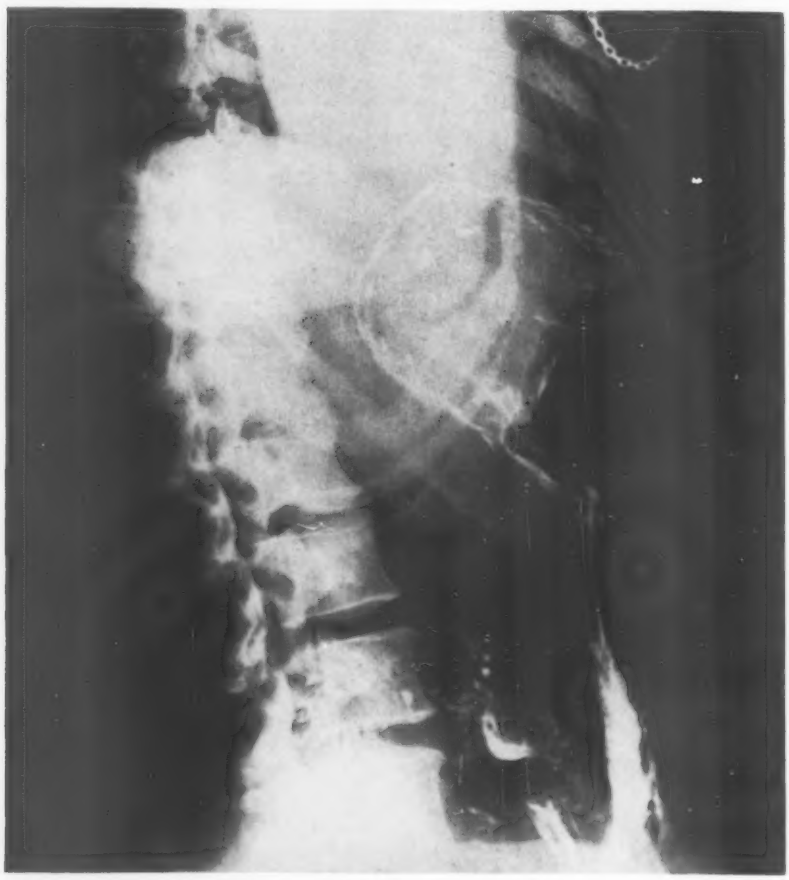


FIG. 3.—Gas in the stomach shows that the cyst is extra gastric which is above and behind the stomach.

half of the spleen was of normal size, shape, color and consistency, but the upper half contained a doughy cyst which measured 9 x 8 x 7 cm. The cyst wall, which was pale in color and had a leathery consistency, appeared to be enclosed within the splenic capsule, but was separated by a portion of the capsule from the splenic pulp. Blood vessels could be demonstrated passing from the spleen to the surface of the cyst. When opened, the cyst was found to be filled with pultaceous material of putty-like consistency. Careful search was made, but no hair, teeth, bone, ecchinococcus ova or hooklets were

found. Wet smear of the cyst contents revealed the presence of large numbers of cholesterol crystals and flakes of dead epithelium. Upon evacuation of the contents of the cyst, the inner wall was found to be atheromatous and had a distinct mammilated appearance.

Subsequent histologic examination of the walls of the cyst revealed thick, hyalinized, fibrous tissue containing areas of bone with well-defined marrow cavities, areas of calcification, and others of necrotic, flaky, material in which there were many cholesterol clefts. The closest approach to an epithelial

FIG. 4.—Removed spleen with cyst.

lining consisted of calcified areas with the configuration of nuclei about which were many cholesterol clefts. Tissue from the boundary between the cyst and splenic pulp revealed thickening of the splenic capsule with some compression of the underlying pulp. Tissue from various areas of the spleen revealed normal splenic structure. The pathologic diagnosis was an epidermoid inclusion in the spleen with cyst formation.

COMMENT

Unlike cysts of the ovary, liver, kidney or other abdominal organs, cysts of the spleen are relatively rare. The first record of any splenic cyst was that

of Andral[1] who discovered the condition in an autopsy in 1829. Pean,[2] in 1867, was the first to remove successfully a splenic cyst. Ecchinococcus cysts of the spleen occur about twice as frequently as various forms of nonparasitic cysts, and false cysts about four times as often as true cysts. Epidermoid cysts, according to Shawan,[3] usually occur in young people of either sex. These cysts, according to Montgomery, *et. al*,[4] are usually large, solitary, and lined with stratified pavement epithelium with prominent intercellular bridges. Custer, as quoted by Montgomery, states that some cysts of this type have been described in which the cavity was filled with sebaceous material and hair and have been reported to weigh as much as 3 Kg., to have contained up to 1,500 cc. of watery, chocolate colored material, and to have contained cholesterol cystals. Fowler[5] pointed out that enucleation of the cyst is rarely feasible and advocates splenectomy as the treatment of choice.

It is interesting to conjecture that our patient's first pregnancy incited her cyst to activity. Fowler reported some association between pregnancy and the incidence of splenic cysts, (quoted by Naidu.[6])

The unusual feature of our case was that the original symptoms were of thoracic origin, the contents having spilled into the pleural cavity.

SUMMARY

A case of an epidermoid cyst of the spleen with symptoms of thoracic origin is reported. Splenectomy was performed through a transthoracic approach. We have found no similar case reported in the literature.

Columbia Hospital
3321 North Maryland
Milwaukee 11, Wisconsin

BIBLIOGRAPHY

1 Andral, G.: Precid d'anatomie pathologique, **2**: 432, 1829.

2 Pean: Gazz. hebd. de med., **4**: 795, 1867.

3 Shawan, H. K.: Epidermoid Cysts of The Spleen. Archives of Surgery, **27**: 63–74, 1933.

4 Montgomery, A. H., Eugene T. McEnery and Albert A. Frank: Epidermoid Cysts of the Spleen. ANNALS OF SURGERY, **108**: 877–844, 1938.

5 Fowler, H. H.: Cystic Tumors, of the Spleen. Int. Abstr. Surg., **70**: 213–223, 1940.

6 Naidu, V. R.: Solitary Splenic Cyst, Indian Medical Gazette, **80**: 131–132, 1945.

EDITORIAL ADDRESS

Original typed manuscripts and illustrations submitted to this Journal should be forwarded prepaid, at the author's risk, to the Chairman of the Editorial Board of the ANNALS OF SURGERY.

John H. Gibbon, Jr., M.D.
1025 Walnut Street, Philadelphia 7, Pa.

Contributions in a foreign language when accepted will be translated and published in English.

Exchanges and Books for Review should be sent to Dr. Gibbon at the above address.

Subscriptions, advertising and all business communications should be addressed

ANNALS OF SURGERY

East Washington Square, Philadelphia, Pa.